Sam Goldstein
Melissa DeVries
Editors

Handbook of DSM-5 Disorders in Children and Adolescents

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This volume is dedicated to the pioneers of mental health research, nosology, and treatment over the past 200 years. Their efforts to create a working, valid, and reliable system of understanding the mental health differences among us are a testament to an ongoing and multi-generational process. This volume is also dedicated to my loving wife Sherrie.

S.G. For Derek and Sevilla. M.D.

Preface

In 1895, Henry Maudsley wrote, "children, because of their immaturity, could not experience mental health problems." He suggested that a mature, grown brain was needed for someone to experience madness. This set the foundation for the widespread belief over the following 100 years that children's brains and their minds were insufficiently developed and unstable, limiting the capacity for them to experience psychopathology. Because of this, few references can be found concerning childhood mental illness or psychopathology prior to the twentieth century. In fact, it was not until the 1930s and the formation of the International Association for Child and Adolescent Psychiatry and Allied Professions that an organized professional group took an interest in children's mental health. Further, it was not until the early 1950s and the founding of the American Academy of Child and Adolescent Psychiatry in 1953 and the European Society for Child and Adolescent Psychiatry in 1954 that child psychiatry and a focus on children's mental health were truly created. Not a single article making reference to children was published in the first 45 years of the *American Journal of Insanity* (1844–1889, forerunner of the *American Journal of Psychiatry*) (Levy, 1968). In fact, Benjamin Rush, noted as the first American psychiatrist, made no mention of children in his textbook *Medical Inquiries and Observations upon the Diseases of the Mind* (1812). The prevailing view was that mental illness did not occur before puberty. Thus, children with mental health problems were simply deemed as misfits or delinquents. Further, puberty was suggested to be a significant cause of insanity (Durand-Fardel, 1885).

The first modern taxonomy of childhood mental disorders was described by Maudsley (1895) in a chapter titled "Insanity of Early Life" (pp. 259–293). Maudsley defined seven different mental health conditions in childhood:

1. *Monomania or partial ideational insanity.* Interestingly, he described these children as possessed by incontrollable impulses.
2. *Choreic delirium or choreic ideational insanity.* Described as children who were extremely aggressive and violent.
3. *Cataleptioid insanity.* Children who demonstrated odd interests, repetitive behaviors, and atypical postures.
4. *Epileptic insanity.* This was thought to be associated with episodes of epilepsy.
5. *Mania.* The condition was not well described.

6. *Melancholia.* Described as a pattern of unhappiness that could lead to suicide.
7. *Affective insanity or moral insanity.* Described as a pattern of "willful wickedness."

Interestingly, considering these early descriptions by Maudsley, one can appreciate their evolution into the current DSM-5 diagnoses. For the most part, these taxonomies were not well anchored in theory. When theory evolved, most psychopathology initially was seen as a form of maladjustment. Symptoms and behaviors were thought to reflect deeply rooted problems within family relationships and the environment. These approaches formed many of the classifications of the earliest versions of the *Diagnostic and Statistical Manual of Mental Disorders* of the American Psychiatric Association.

As far as we are aware, this volume represents the first volume specifically devoted to DSM disorders in childhood. Since its first publication, as a small diagnostic manual in 1952, the DSM list of diagnoses, diagnostic criteria, and supporting information has grown into manuals consisting of hundreds and hundreds of pages. We have organized this volume about the DSM-5 in childhood, specifically focusing on the internalizing/externalizing framework first described by John Werry (1985). We have edited this handbook as a desk reference for mental health professionals, providing in-depth information and guidance extending beyond the brief descriptions in DSM-5. We have also included a number of chapters focusing on psychosocial treatments for many of these conditions.

We very much appreciate the efforts and diligence of our contributing authors. It is our hope and intent that this volume will serve as an important resource in childhood mental health, the diagnostic process, and, most importantly, treatment.

Salt Lake City, UT, USA — Sam Goldstein
March 2017 — Melissa DeVries

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Quotes

A desire to be in charge of our own lives, a need for control, is born in each of us. It is essential to our mental health, and our success, that we take control.

Robert Foster Bennett

Mental health needs a great deal of attention. It's the final taboo and it needs to be faced and dealt with.

Adam Ant

We take our kids for physical vaccinations, dental exams, eye check-ups. When do we think to take our son or daughter for a mental health check-up?

Gordon Smith

Contents

About the Editors

Sam Goldstein, PhD is a doctoral-level psychologist with areas of study in school psychology, child development, and neuropsychology. He is licensed as a psychologist and certified as a developmental disabilities evaluator in the state of Utah. Dr. Goldstein is a fellow in the National Academy of Neuropsychology and American Academy for Cerebral Palsy and Developmental Medicine. Dr. Goldstein is an assistant clinical instructor in the Department of Psychiatry. Since 1980, Dr. Goldstein has worked in a private practice setting as the director of a multidisciplinary team, providing evaluation, case management, and treatment services for children and adults with histories of neurological disease and trauma, learning disability, adjustment difficulties, and attention deficit disorder. Dr. Goldstein is on staff at the University Neuropsychiatric Institute. He has served as a member of the Children's Hospital Craniofacial Team. He has also been a member of the Developmental Disabilities Clinic in the Department of Psychiatry at the University of Utah Medical School.

Dr. Goldstein has authored, co-authored, or edited 38 clinical and trade publications, including 17 textbooks dealing with managing children's behavior in the classroom, genetics, attention disorder, and adult learning disabilities. With Barbara Ingersoll, Ph.D., he has co-authored texts dealing with controversial treatments for children's learning and attention problems and childhood depression. With Anne Teeter Ellison, he has authored *Clinician's Guide to Adult ADHD: Assessment and Intervention*. With Nancy Mather, Ph.D., he has completed three texts for teachers and parents concerning behavioral and educational issues. With Michael Goldstein, M.D., he has completed two texts on attention deficit hyperactivity disorder. He has edited three texts with Cecil Reynolds, Ph.D., on neurodevelopmental and genetic disorders in children. With Robert Brooks, Ph.D., he has authored 11 texts including the *Handbook of Resilience in Children*, *Understanding and Managing Children's Classroom Behavior – 2nd Edition*, *Raising Resilient Children*, *Nurturing Resilience in Our Children*, *Seven Steps to Help Children Worry Less*, *Seven Steps to Anger Management*, *The Power of Resilience*, *Raising a Self-Disciplined Child*, and *Raising Resilient Children with Autism Spectrum Disorders*. With Jack Naglieri and Sally Ozonoff, Ph.D., he has authored a number of texts on autism and assessment of intelligence and executive functioning. He has co-authored a parent training program and is currently completing a number of additional texts on resilience, ADHD, and genetics. Dr. Goldstein is the editor in chief of the *Journal of Attention Disorders* and serves on seven editorial boards. He is also the co-editor of the

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Dr. Goldstein, a knowledgeable and entertaining speaker, has lectured extensively on a national and international basis to thousands of professionals and parents concerning attention disorders in children, resilience, depression, adjustment and developmental impairments, autism, and assessment of brain dysfunction.

Melissa DeVries, PhD is a doctoral-level psychologist with areas of study in school psychology, neuropsychology, and child development. She is a licensed psychologist and is a member of the National Association of School Psychologists and the American Psychological Association. Dr. DeVries has worked at the Neurology Learning and Behavior Center in Salt Lake City, Utah, since 2009 providing evaluation, case management, and treatment services for children and adults with developmental disorders, mood and anxiety disorders, post-traumatic stress, and learning disorders. She also provides school psychology consultation services to the Montessori Community School. Dr. DeVries has co-edited a textbook on learning and attention disorders and has also co-authored several chapters on these topics as well as genetic syndromes, executive functioning, and resilience. Dr. DeVries is also an editorial board member for the *Journal of Attention Disorders*.

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Part I

Introduction

Diagnosis in American Psychiatry: A Brief History of the Diagnostic and Statistical Manual

1

Faisal Akram, Shadia Kawa, and James Giordano

Introduction

> Classification is the formal human process analogous to concept formation that occurs in all higher animals as they attempt to master their environment. (Spitzer & Wilson, 1975)

Nosology, from the Greek *nosos* (disease) and *logos* (rational study), is the discipline and practice(s) that deal with the classification of disease. Nosology has been a focus of attention throughout the history of psychiatry. The current, fifth edition of the American Psychiatric Association's *Diagnostic and Statistical Manual* (*DSM-5*) has, like prior editions of the work, become something of a benchmark for standardizing classification of psychiatric disorders. Reflecting this hegemony, the *DSM-5* is viewed as the authoritative resource for assessment and characterization of mental disorders, for use by clinicians, researchers, policy makers, insurance agencies, and legal systems ("American Psychiatric Association [APA]," 2014). A major feature of the *DSM*, and *DSM-5* in particular, is homogenization and universalization of psychiatric diagnoses, which has become somewhat normative in psychiatric practice, not only in the United States but increasingly in Europe and Asia, given current trends toward concomitantly employing *DSM* and International Classification of Diseases (ICD) standards and criteria (Demazeux & Singy, 2015).

Yet, each successive revision of the *DSM* has generated increasing scholarly discourse and criticism. In many ways, the ongoing discourse – and debates – about the basis and meanings of psychiatric nosology and its effect upon medical practice as well as social constructs (of normality and abnormality) reflect the history of psychiatry writ large. This historicity provides insight to iterative changes in psychiatry as a discipline and set of practices, the role of biomedicine and technology in medicine, and changing social values and stances. In this light, we believe that a historical overview may shed important light upon contexts of iterative developments and modifications in psychiatric nosology – and practice(s) – within an evolving scientific, medical, and sociopolitical milieu. In this chapter, we trace the roots of current discourses – and controversies – starting from nineteenth-century psychiatry to the publication of the *DSM-5*, with some speculation as to what the *DSM* may portend for the near future. At this point, it may be worth mentioning what Micale (1996) has called the "paradigmatic structuring of psychiatric historioriographies," the unintentional but implicitly biasing phenomenon of emphasizing theories, figures, and facts of a prevailing theoretical paradigm (e.g., the biomedical

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orientation) and marginalizing competing ideologies and facts. Acknowledging these tendencies, we hope to maintain an impartial perspective on this brief recollection of events which led to the development of *DSM-5*.

The Beginnings of Psychiatric Nosology

In the nineteenth century, the practice of psychiatry was conducted either in (rather posh) private office settings for those patients who were of upper social standing or in asylums for those patients of more proletariat or impoverished social standing. Of additional note and import is that mental illness was commonly referred to as insanity. Some of the earliest attempts to classify insanity can be seen in a treatise by Philippe Pinel and his foremost pupil Jean-Étienne Dominique Esquirol (Esquirol, 1845; Pinel, 1806). In the United States, a crude form of classification of insanity into mania, melancholia, dementia, and idiocy is found in 1838–1844 Annual Reports of the Trustees of State Lunatic Hospital at Worcester (Worcester, 1838, 1839, 1840, 1841, 1842, 1843, 1844). However, the 1840 report noted:

> The division of insanity into these four classes is somewhat arbitrary. In the strongly marked cases, the distinctions are very plain, no less so by natural language than by symptoms of disease; as they approximate each other, the difficulty of classification is increased till it is found impossible where the cases shall be arranged.

Germane to our theme, these reports also discuss the nature of insanity at some length. For nineteenth-century asylum psychiatrists, insanity was a physical disease of the brain, yet it was mysteriously suborned by immaterial entities like immorality, stress, and psyche (Grob, 1962, 1991). To be sure, nineteenth-century nosological attempts in the United States were not intended for diagnostic clarity; rather, they were a quest for empirical data to inform social policies concerning the institutionalized mentally ill (Grob, 1991). These nosological efforts reached their peak in the progressive era (1890–1914), when the roles of census and statistics grew stronger in social and scientific disciplines (Gould, 2001). By 1917, the Bureau of Census and the American Medico-Psychological Association (what is now the American Psychiatric Association) had already conjoined efforts to develop a uniform nomenclature that could serve to expedite the collection of statistical data on mental illness.

In Europe, Emil Kraepelin had developed a system of classifying psychiatric conditions, which, contrary to intentions behind nosological attempts in the United States, were primarily aimed at revealing the "secrets of nature" by operationalizing psychiatric symptoms and observing the natural course of illness (Hippius & Müller, 2008). Kraepelin's teachings were communicated to members of the American Medico-Psychological Association through his textbooks, editions of which were anticipated with same degree of anticipation (and in some cases vehemence) as would be later observed with editions of the *DSM*. Subsequently, motivated by the Bureau of Census and inspired by Kraepelin's work, the American Medico-Psychological Association (with cooperation of the National Committee for Mental Hygiene) issued the first standardized psychiatry nosology, *Statistical Manual for the Use of Institutions for the Insane*, in 1918. This manual had 22 categories of disorder (predominantly psychoses), of which 20 were based on Kraepelinian constructs. Nine subsequent editions of the *Statistical Manual* followed during the interwar years. None were endorsed by psychiatrists; after all, diagnosis was not essential to treatment at that time. Instead, psychiatrists were attending to Freudian conceptualizations of disorder as derived from and based upon psychoanalytic theory. Nevertheless, the *Statistical Manual* established a niche as defining American psychiatric nosology in the period prior to the Second World War.

DSM-I Development and the Post-*DSM-I* Era

During (and after) World War II, Freudian psychoanalysis, promoted by Meyerian "search for life events"(McHugh, 2016), proved to be relatively successful in treating "combat neurosis"

and other "psychoneuroses"(Grob, 1987). As a result, psychodynamic theories gained rapid acceptance in military psychiatry, so much so that William C. Menninger, director of the Psychiatry Consultants Division of the United States Army, developed an independent nomenclature called "War Department Technical Bulletin, Medical 203," which was predominantly psychodynamic in orientation. Psychodynamic theories challenged both Kraepelinian doctrine and, albeit more indirectly, the traditional medical model of disease. When military psychiatrists returned home after the war, they further reinforced psychodynamic trends and psychoanalytic traditions and, in so doing, adopted roles as reformers of American psychiatry.

The conceptualization of psychopathology largely shifted from recognizing mental conditions as discrete disease entities that were distinct from mental health to considering mental health and illness on a continuum of variable severity. Reflecting upon shifting constructs of mental disease, and a growing population of psychiatric patients, the APA Committee on Nomenclature and Statistics sought a new system of classification (Raines, 1953). Led by George Raines, a former United States Navy neuropsychiatrist and then professor of psychiatry at Georgetown University Medical Center, the first edition of the *Diagnostic and Statistical Manual of Mental Disorders* was released in 1952 (APA, 1952).

The *DSM-I* can be regarded as official evidence of the growing preeminence of psychoanalysis in American psychiatry, as it classified mental disorders into two major groups: (1) conditions that were presumed to be caused by organic brain dysfunction (e.g., as associated with trauma, intoxication, or somatic diseases) and (2) conditions that presumably resulted from socio-environmental stress and its impact upon patients' constitution and adaptive ability. While considered to be an important step in the evolution of psychiatric nosology, the *DSM-I* actually exerted little to no influence on psychiatric practice (Braslow, 2000). In fact, normality, neuroses, and psychoses were considered to be on the same continuum in psychodynamic psychiatry; therefore, diagnosis was considered to be irrelevant. What mattered was early intervention, which could impede the progression of psychological dysfunction to severe psychoses (Menninger, 1964).

Many psychiatrists sought to intervene at a social level, as it was viewed as the source of actions (socio-environmental factors) that triggered "reactions" (a term considered to be synonymous with mental illness in the Meyerian tradition). As a consequence, psychiatry adopted a type of social responsibility (Group for the Advancement of Psychiatry, 1950) and mental health policies shifted in favor of this approach (Grob, 1987). Highly influential, the "Group for the Advancement of Psychiatry" (GAP) further promoted this social activism through its reports, which applied psychiatry to broad social issues such as child desegregation, industry, civil defense, and international relations (GAP, 1950, 1951, 1957).

By the 1960s, the legitimacy of "asylum psychiatry" was clearly in question, and "community psychiatry," driven by psychoanalysis and social activism, tended to be ever more representative of both the professional stance and "public face" of the contemporary psychiatry. Indeed, psychoanalysis reached the community and was very well received by general public. Dynamic psychiatrists were considered – and portrayed – as purveyors of reason, knowledge, and well-being (Gabbard & Gabbard, 1999). Psychiatry became delimited from traditional diagnostics to engage a wider community in which diverse problems such as failing marriage, troubled childhood, and personal dissatisfaction were now seen as viable for clinical intervention (Hale Jr, 1995). In short, psychiatry came closer to being established as both a charismatic medical specialty and a growingly forceful social discipline. Yet, as Grob (2011) has noted, "… at precisely the time that the social legitimacy of psychiatry peaked, a series of disquieting elements were already eroding its very foundation."

DSM-III: A Nosological Revolution

While a shift in the conceptualization of mental illness radically transformed American psychiatry, it also sparked a number of controversies during the 1960s and 1970s (Grob, 1987). The fluidity of construct boundaries between what constituted mental health and mental illness, inherent to psychodynamic concepts of mental illness, became the source of two extremely different perspectives on psychiatry. On the one hand, psychiatry activists attributed every wrongdoing of society to mental illness, while on the other, Szasz (1961) and several others (Goffman, 1961; Scheff, 1970) argued that psychiatry was merely a discipline for social control of unwanted behavior, as there was no "true mental illness"(Szasz, 1961). They thus challenged the medical legitimacy of psychiatry for the first time in the history of the field. Szasz's argument was further strengthened by the fact that psychodynamic therapy services, most of which had not been rigorously investigated, were at that time being offered not only by psychiatrists and clinical psychologists but by non-medically licensed professionals (e.g., social workers and counselors; Mayes & Horwitz, 2005). Moreover, the deinstitutionalization movement liberated many patients with chronic mental illnesses from mental hospitals and allowed them to freely enter into the community. Psychoanalytic therapies, once tremendously successful in treating "psychoneuroses," failed to deal with serious and debilitating mental illness.

Instead, by the 1960s the use of drugs – psychopharmacology – had become ever more en vogue, fortified, in due part, to the development of a significant armamentarium of mood- and behavior-modifying agents, which proved to be effective in alleviating psychiatric symptoms and restoring daily functioning of mentally ill patients (Ban, 2001). This "psychopharmacological turn" fostered the rise of an "invisible college" of biologically oriented psychiatrists who were more neo-Kraepelinian in focus and critical of psychodynamically oriented approaches (Millon & Klerman, 1986).

When the second edition of the *Diagnostic and Statistical Manual of Mental Disorders* was published in 1968 (APA, 1968), psychodynamic approach was already losing its hegemony on psychiatry. Although the *DSM-II* was generally aligned with Freudian traditions, subtle amendments hinted at change. For example, there was a notable decrease in the use of the psychodynamic term "reaction." A disclaimer accompanied this development:

> Some … may interpret this change as a return to a Kraepelinian way of thinking, which views mental disorders as fixed disease entities. Actually this was not the intent of the APA Committee on Nomenclature and Statistics: "[The Committee] tried to avoid terms … regarding either the nature of a disorder or its causes [...]. In the case of diagnostic categories about which there is current controversy … the Committee has attempted to select terms which it thought would least bind the judgment of the user.

The total number of disorders increased from 106 in *DSM-I* to 182 in *DSM-II*, with inclusion of milder disturbances such as Conditions without Manifest Psychiatric Disorder and Transient Situational Disturbances, suggesting evidence of a natural progress in classification of mental illnesses. The *DSM-II* Task Force also made first attempts to classify childhood psychopathology based on psychodynamic concepts; included were now a separate subgroup Behavior Disorders of Childhood and Adolescence containing seven "reactions": hyperkinetic, withdrawing, overanxious, runaway, unsocialized aggressive, group delinquent, and other reactions of childhood (or adolescence). It is worth mentioning here that prior to the release of the *DSM-II*, the GAP had already proposed an independent classification system for psychopathological disorders in childhood. GAP's proposed classification had a typically psychodynamic flavor and divided behaviors – presumably on a continuum – into healthy responses, developmental deviations, and disorder categories.

However, the reliability of both classification systems was relatively low (Achenbach & Edelbrock, 1978; Beitchman, Dielman, Landis, Benson, & Kemp, 1978; Freeman, 1971). This issue of subpar reliability not only was limited to

childhood mental disorders but rather was problematic for all psychiatric diagnoses (Sandifer, Pettus, & Quade, 1964; Tarter, Templer, & Hardy, 1975). Diagnosis, in turn, had by then become relevant – and a matter of concern – to psychiatrists because it had become the basis for prescribing medications, insurance reimbursements, clinical research trials, and research grants (Mayes & Horwitz, 2005; Wilson, 1993).

With progress in biological psychiatry and psychopharmacology (Baer, Platman, & Fieve, 1970; Klerman, 1968; Sachar, 1970), negative critique of the extant psychiatric nosology mounted considerably during the 1960s and early to mid-1970s. Moreover, significant improvements in quantitative psychiatric assessments, through symptom-based rating scales, raised the possibility of defining a mental disorder using a descriptive phenomenology approach. In fact, Lorr, Sonn, and Katz, (1967) had called for a phenomenological definition of depression prior to the release of *DSM-II* (Lorr et al. 1967). In 1972, the Washington University group, under the supervision of Eli Robins and Samuel Guze, demonstrated how a phenomenological approach could be utilized to operationalize mental symptoms, and proposed the "Washington University Diagnostic Criteria" for the diagnosis of mental disorders (Feighner et al., 1972). This seminal work established the Foundation for Research Diagnostic Criteria (Spitzer, Endicott, & Robins, 1978) and, in so doing, created a propitious climate for change that culminated in the publication of the third edition of the *Diagnostic and Statistical Manual of Mental Disorders* in 1980 (APA, 1980).

The publication of the *DSM-III* was, by any measure, a nosological revolution as the classifying paradigm shifted from being etiological/phenomenological to being more purely phenomenological. The *DSM-III* Nomenclature Task Force incorporated a polythetic criteria-based categorical system of classification for the first time in the history of psychiatric nosology. Such delicate attention to symptoms and their reorganization into categories heralded a return to Kraepelinian traditions. Perhaps the most telling feature of this trend was the official removal of the psychodynamic term "neurosis" in the draft, which, after waves of protest, would later appear only parenthetically as neurotic disorder. Neuroses were renamed and placed under several categories including affective, anxiety, somatoform, and dissociative disorders.

The increased number of mental disorder categories (from 182 in the *DSM-II* to 265 in the *DSM-III*) was partly reflective of the increase in psychiatric knowledge achieved since the publication of the *DSM-II* and was also a consequence of gross reorganization of previously broad categories into several individual subcategories or subtypes, each considered as a separate and discrete mental disorder. For instance, *DSM-III* introduced an Axis I category "Disorders Usually First Evident in Infancy, Childhood, and Adolescence," which included mental retardation; conduct disorder (with five subtypes); attention deficit disorder (with and without hyperactivity); four specific eating disorders (previously a single category "Feeding Disturbance" in *DSM-II*); anxiety disorders (separation anxiety, avoidant and overanxious disorders); three stereotype movement disorders (previously a single category, Tic, in *DSM-II*); pervasive developmental disorder (including infantile autism); five other disorders of infancy, childhood, or adolescence (including oppositional, schizoid, and identity disorders); and five other disorders with physical manifestations (stuttering, enuresis, encopresis, sleepwalking, and sleep terror disorders). Similarly, specific learning disturbance of *DSM-II* was divided into five different specific developmental disorders. In addition, many novel disorder categories such as bipolar disorder, post-traumatic stress disorder, psychosexual dysfunctions, and four disorders of impulse control not elsewhere classified were also incorporated. Further, as based on the consensus opinion that the earlier classifications of homosexuality as disorder were largely shaped by politically and socioculturally contingent notions of deviance rather than scientific corroboration, homosexuality was permanently removed from psychiatric nomenclature and nosology (Millon & Klerman, 1986).

But the main innovation in the *DSM-III* was not in the renaming and reorganization of the disorder categories, but in the articulation of explicitly defined operational criteria, and listing of symptoms and durations a patient would require in order to be given a particular diagnosis. Another unique feature of the *DSM-III* was the adoption of a "multiaxial system" of diagnosis that emphasized the interaction of presenting symptoms, personality, physical health, psychosocial and environmental factors, and functioning in daily life. One development that we feel deserves special mention was the field testing of *DSM-III* diagnoses through National Institute of Mental Health (NIMH)-sponsored trials before publication. Although it was not compared to inter-rater agreement(s) of earlier nosologies, the *DSM-III* classification system demonstrated relatively good diagnostic reliability (Spitzer et al., 1978) and was officially released in 1980.

The Post-*DSM-III* Era: Transformation of American Psychiatry

Soon after its release, Gerald Klerman, a leading psychiatrist, labeled the publication of *DSM-III* as "...another major turning point similar to the acceptance of chlorpromazine" (Klerman, 1984). Within 6 months of its publication, more orders were received for the *DSM-III* than all the previous *DSM* editions combined, including their 30-plus reprintings (Mayes & Horwitz, 2005). Effects of *DSM-III* were evident in all dimensions of psychiatry including research, clinical practice, and education. For clinicians, *DSM-III* proved their claim that they were treating "true illnesses." For insurance providers, it provided standards for reimbursement of therapeutic modalities. For medical students and psychiatry residents, it became "*the* reference" for diagnosis, if not conceptualization of mental disorders, to the extent that American medical schools and residency programs expected students and physicians to pass examinations based on *DSM-III* criteria (Young, 1997).

In retrospect, the *DSM-III* marked the beginning of the end of Freudian influence on American psychiatry. While a defense of conventional medical model of disease was not explicitly stated in *DSM-III*, the process of its development made it clearly implicit. In fact, the decision of the *DSM* Task Force to maintain a stance of being "atheoretical" about the nature of mental disorder with diversion of research interests (and funding) to neuroscience, neurobiology, and psychopharmacology was reflective of a general belief among the research community that the biomedical (i.e., physical) nature of mental disorders will be eventually proven. In this light, the *DSM-III* achieved tremendous success in establishing the medical legitimacy of psychiatry (Pasnau, 1987). Several epidemiological studies, including the NIMH Epidemiological Catchment Area Project, followed to assess the incidence and prevalence of mental disorders.

The landscape of psychiatric research changed completely as all scientific inquiries in psychiatry were heretofore directed at *DSM-III* disorders. For psychopharmacology, the *DSM-III* catalyzed the dawn of a new era of randomized controlled trials (RCTs) directed at specific disorders. In the following years, billions of dollars were allocated by the government and pharmaceutical companies for psychopharmacological research. Psychopharmacological agents became the first line of treatment for the majority of mental disorders. The culmination, if not fruition of this progress, was evidenced in the US Congressional declaration that the 1990s were to be the "Decade of the Brain".

DSM-IIIR, *DSM-IV*, and the Rising Tides of Controversy

Subsequent editions of the *DSM* were generally in keeping with the scope and tenor of the *DSM-III*. An updated edition, the *DSM-IIIR*, was published in 1987 (APA, 1987) and incurred minor revisions such as the changing of attention deficit disorder to attention deficit hyperactivity disorder, placement of mental retardation and pervasive development disorder in Axis II category, deletion of schizoid disorder of childhood (or adolescence), and the renaming of affective disorders as mood disorders. Similarly, *DSM-IV*

and the text-revised edition, *DSM-IV-TR*, published in 1994 and 2000, respectively (APA, 1994, 2000), reflected minor additions, deletions, and reorganization of diagnostic categories of the descriptive phenomenological approach initially adopted in the *DSM-III*.

With each successive edition of the *DSM*, the specificity of operational criteria and, in turn, the total number of diagnoses increased. For instance, the diagnostic criteria for autistic disorder were substantially revised, and two diagnoses, Rett's disorder and Asperger's disorder, were added to the pervasive developmental disorder group. Similarly, avoidant disorder and overanxious disorder of childhood were eliminated because their diagnostic criteria were not specific and resembled those of social phobia and generalized anxiety disorder (Shaffer et al., 1996). In addition, acknowledging cultural variability in the ways that mental health and illness are expressed and construed, "culture-bound syndromes" were also included in the *DSM-IV* and *DSM-IV-TR*.

A major development in transition from *DSM-IIIR* to *DSM-IV* was the inclusion of the "clinical significance" criterion to most of the categories of mental disorder. Inclusion of this criterion was a response to increasing criticism from the psychiatric community that the *DSM-IIIR* approach had a tendency toward "medicalization" of normal behavior. Most notable was the evidence provided by Shaffer et al. (1996) that there were many children and teenagers who met *DSM-III-R* criteria for diagnoses, but who did not appear to have significant social or academic impairment, and had never been referred for any clinical service (Shaffer et al., 1996). In reality, the heart of this issue was the same fluidity of the boundary between mental health and mental illness, which, after the loss of psychodynamic hegemony, was now resurfacing in the post-*DSM-III* era to raise controversial issues. Thus, the concept of mental disorder had to be reevaluated.

Proposed that "disorder is a harmful dysfunction, wherein 'harmful' is a value term based on social norms, and dysfunction is a scientific term referring to the failure of mental mechanism to perform a natural function for which it was designed by evolution." To some extent, inclusion of the "clinical significance" criterion conformed to this "harmful dysfunction" analysis; however, it was not applied to all mental disorders and was even reversed for Tic disorder in the *DSM-IV-TR* (First & Pincus, 2002). The lack of clear demarcation between mental health and illness raised other issues, as well, inclusive of problems with under- and/or overdiagnosis and the "pharmacologization of psychiatry" (Timimi, 2014).

The classifying approach for diagnosis of developmental disorder was also questioned (Cohen, Paul, & Volkmar, 1986). In particular, the deficiency of a systematic, multidisciplinary, developmentally based classifying approach in the *DSM* was immediately felt in infant and early childhood mental health practice, which led to the development of the *Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood (DC:0-3)* (Wieder, 1994). The effect was noted even in the psychiatric care of older children, as there was an evident surge in pediatric bipolar disorder diagnoses (Frances, 2010; Parry et al., 2008; Zimmerman, Ruggero, Chelminski, & Young, 2008). Parallel to this broadening categorization of mental disorders was a consistent increase in the use of psychopharmacological interventions. This was – and continues to be – heavily criticized, especially in relation to the risk of suicide consequential to (outpatient) antidepressant use (Hammad, Laughren, & Racoosin, 2006) and use of psychostimulants for cognitive performance enhancement (del Carmen Panini, Garraza, Teves, Giraudo, & Calderón, 2015; Insel, 2009; Moynihan & Cassels, 2006).

The Development of the *DSM-5*

The process of *DSM-5* development began in 1999, and a major concern was to address a range of issues that had emerged over the previous 30 years of *DSM* use in practice. Nowhere were these issues more accurately depicted than in *A Research Agenda for DSM-V* (Kupfer, First, & Regier, 2008). Briefly, these issues were related to: (1) the definition of a mental disorder,

(2) relationships between mental disorder and disability, (3) debates about adding dimensional criteria to diagnoses, (4) potential addition of a neurodevelopmental perspective to nosology to account for variable manifestations of an illness across developmental stages of the life span, and (5) incorporation of neuroscience research-based data in diagnostic criteria.

Perhaps the most pressing issue was the increasing gap between insights gained by neuroscience research and their application to psychiatric practice. The Decade of the Brain evoked a significant growth in neuroscience research. Such research highlighted interesting aspects of *DSM* classification which led many to then question the validity of *DSM*-based diagnoses (Kendell & Jablensky, 2003; Lahey et al., 1998; Morgan, Hynd, Riccio, & Hall, 1996). To wit, David Kupfer, chair of the *DSM-5* planning committee, together with coauthors Michael B. First and Darrel E. Regier, stated:

> Those of us who have worked for several decades to improve the reliability of our diagnostic criteria are now searching for new approaches to an understanding of etiological and pathophysiological mechanisms— an understanding that can improve the validity of our diagnoses and the consequent power of our preventive and treatment interventions. (Kupfer et al., 2008)

The issue of significant comorbidity and concurrent overlap of neuroscience research findings for distinct *DSM* disorders remained problematic throughout the development of *DSM-5* (Berrettini, 2000; Doherty & Owen, 2014; Kendler, Neale, Kessler, Heath, & Eaves, 1992). This overlap of research findings implied that there would not be enough evidence-based data to reliably differentiate between what had been posited to be nosologically discrete psychiatric disorders. As a result, the opportunity to incorporate neurobiological data from neurogenetics, structural and functional neuroimaging, psychophysiology, developmental psychopathology, and pharmacogenomics was lost, and, against the hopes of the majority of psychiatric community who were envisioning the *DSM-5* as a revolutionary neuroscientifically based classification system, the volume was published in 2013, as what appeared to many to be little more than a revised, descriptive phenomenologically based nosology (APA, 2013).

Yet, changes in the *DSM-5* deserve a mention. The total number of categories increased from 297 in *DSM-IV-TR* to 541 in *DSM-5*; however, unlike previous editions, the number of defined diagnoses decreased from 172 in *DSM-IV-TR* to 157 in *DSM-5*. Disorders were reordered into a revised organizational structure, with disorders more frequently diagnosed in childhood (e.g., neurodevelopmental disorders) at the beginning of the manual and disorders more applicable to older adulthood (e.g., neurocognitive disorders) at the end. Introduction of a new organizational structure was in recognition of a model of internalization-externalization that has gained increasing support over the years (Krueger, 1999; Krueger & Markon, 2006).

Age, gender, and culturally related factors that are specific to diagnosis were included. The *DSM-IV* "Not Otherwise Specified" category was further divided into "Other Specified" and "Unspecified" categories. The *DSM-IV* subtypes of schizophrenia (i.e., paranoid, disorganized, catatonic, undifferentiated, and residual types) were eliminated, and autistic disorder, Asperger's disorder, and pervasive developmental disorder were placed on a single spectrum under the umbra of autism spectrum disorder after elimination of disintegrative disorder and Rett's syndrome. Individual disorders, diagnostic categories, and criteria were revised to better serve young patients. Social communication disorder (SCD), disruptive mood dysregulation disorder (DMDD), and post-traumatic stress disorder for children 6 years and younger were newly included. A unique feature of the *DSM-5* was the addition of a new section (Section III) to highlight disorders that required further study, but were not sufficiently well established to be a part of the official classification of mental disorders for routine clinical use. Finally, an alternative, "hybrid" model of classifying and conceptualizing personality disorders was also a feature of Section III, which discussed the possibility of integrating categorical and dimensional models for better clinical utility.

Moving Beyond the *DSM-5*

Overall, *DSM-5* did not meet the expectations of the psychiatric community. Controversial issues surrounding the *DSM-IV* continued to haunt the *DSM-5* as well. Comorbidities of disorders are likely to remain high. The concept of mental disorder is vague and diagnostic uncertainty prevails. Yet, most people who go to a psychiatrist get a diagnostic label, be it in the "Other Specified" or "Unspecified" category, which invokes criticisms such as "pathologization of deviance" and the "medicalization of social ills" on a regular basis. Similarly, discourse – if not controversy – continues about the interpretation of subjective variables, such as what constitutes "normal" or "optimal" function within the context and expectations of society and culture (Giordano, 2014; Nagel, 2014).

Perhaps the most disappointing aspect of the *DSM-5* has been the lack of neurobiological data in establishing diagnostic criteria. Two parallel views have emerged to explain why neuroscientific research has failed to inform psychiatric nosology. One view attributes this to the inherent complexity of the brain, persistent unknowns regarding the nature of consciousness and brain-mind relations, and unrequited hopes that neurobiological data will validate *DSM* diagnoses (Nemeroff et al., 2013); the other holds biblical adoption of *DSM* diagnoses as being responsible for the continuing negligibility of neuroscientific information on and in psychiatric practice (Cuthbert, 2014; Cuthbert & Insel, 2013).

Arguably, standardized diagnostic classifications, based on conceptualization of discrete disease entities, can legitimize existence of named and defined disease(s), and in so doing both reduce (at least the overt appearance of) social constructivism and sustain ontologic claims of medicine (Rosenberg, 2002; Patil and Giordano, 2010). Hence, it may be worth pondering the extent to which such a phenomenon could be problematic in psychiatric research. Most neuroscientific research in the post-*DSM-III* era was intended to examine *DSM* diagnoses as if they were "natural disease entities," and in so doing, the mixed monothetic/polythetic criteria-based categorical approaches inherent to *DSM*-based diagnoses invariably incorporated considerable comorbidity and symptom heterogeneity in study populations. This produced similar degrees of variability in results (even for the most sophisticated assessment techniques of neuroscience), thereby decreasing their reliability – if not validity in certain instances. As a result, a vast majority of neuroscience research findings are of moderate or small effect sizes and attempts to replicate these findings have failed for the most part (Kapur, Phillips, & Insel, 2012).

To counter such problems, the NIMH initiated the Research Domain Criteria (RDoC) project in 2009 so as to liberate researchers from current diagnostic nosology of the *DSM* and ICD systems. The RDoC shift the focus of psychiatric research to identify biologically homogenous subtypes that extend across phenotypic diagnoses, rather than validating traditional *DSM* diagnoses, and in this way, aims to enable a more contributory engagement of neuroscientific research in an integrative science of psychopathology (Kozak & Cuthbert, 2016). The knowledge base acquired from RDoC-oriented research will transform psychiatric nosology (Cuthbert, 2015; Cuthbert & Insel, 2013) to a dimensional model of classifying psychopathology, integrating several levels of information (e.g., epigenetics, neurogenomics, structural and functional neuroimaging, neurophysiology, molecular psychiatry, and psychology) to validate and conceptually refine conceptualizations of cognitions, emotion, and behavior. Moreover, the RDoC framework has increasingly emphasized the temporal interaction of neurological development and the environment in the formation of personality, behavior, and mental illness (Casey, Oliveri, & Insel, 2014).

Although still rather new, the RDoC project represents a viable approach to addressing many problematic issues currently arising from the use of the *DSM-5* and *DSM*-based diagnostic criteria. The use of dimensional rather than categorical or diagnostic measures may provide greater traction for discovery of brain-behavior associations. Specifically, with the framework intentionally constructed to increasingly incorporate iterative

findings from neuroscientific research, the prospects of utilizing neurotechnology for the assessment and characterization of mental disorders appear brighter, and consequently, both reliability and validity of diagnosing mental disorders are expected to increase. Moreover, the neurodevelopmental and environmental emphases of the RDoC framework are better suited to address the challenges faced when assessing young children, in whom rapid developmental changes, immediate relevance of a dynamic caregiving environment, and temperamental variations are of considerable interest and importance.

However, some issues of psychiatric nosology are likely to persist even after adoption of a neuroscience-based classification system. For example, in light of recent findings from neuroscience research, human cognition, emotion, and behavior are increasingly being conceptualized to function as continuous variables, which make demarcation of what is ordered or disordered a value judgment, which would be based on the context and expectations of society. Therefore, issues such as "distinction between normality and abnormality" and "pathologization of deviant behavior" could become even more problematic. Similarly, the search for biomarkers, biotypes, and endophenotypes to serve as diagnostic criteria of psychiatric disorders is likely to (continue to) raise concerns about stigmatization and over-medicalization (Singh & Rose, 2009).

There is also continuing focus upon – and debate about – what constitutes treatment or enhancement, what measures should be applied to define and assess these characterizations, and what guidelines and policies should be developed and employed in accordance with these criteria and to direct and govern psychiatric interventions toward such practices (Giordano & Shook, 2015; Shook & Giordano, 2016). Perhaps, solutions to these issues reside in recognizing ideologies, values, standards, and contexts that influence (or, in some cases, define) those ways that scientific knowledge is acquired and employed. Such recognition will require interdisciplinary engagement of neuroscience researchers, psychiatrists, social scientists, ethicists, legal scholars, and policymakers and will likely remain a work in progress as psychiatric classifications are proposed, established, and employed in research, clinical practice, and the social realm.

Conclusion

The history of the *DSM* may certainly be viewed as an attempt to utilize the existing epistemological capital of "science" and "medicine" in the classification of psychopathology. But, as Almeder (1998) has noted, considerations and concepts of science and medicine are often mutable and vary with types and level of knowledge available, cultural influence, and changes in regnant ideas and schools of thought. Each iteration of the *DSM* has provided its own framework for conceptualization and inquiry of mental illness to develop a knowledge base for future nosology. It is important to realize that current psychiatric diagnoses are, to some extent, "constructed entities" that will undergo continual refinement and modification, until such time that they are able to stand as irrefutable, and "carve nature at its joints." Many of the controversies surrounding the *DSM-5* may be related to the concept of "disorder." As such, we posit that it would be wise to consider "disorder" in Karl Jaspers' terms as "an objective which one cannot reach since it is unending…" but which "… indicates the path for fruitful research and supplies a valid point of orientation for particular empirical investigations." The evolution of the *DSM* illustrates that values can and often do play a crucial role in developing nosology. According to Sadler, Hulgus, and Agich (1994):

> …values… determine what we select as 'important,' 'crucial,' 'central,' 'decisive,' or 'related' … consequently, 'descriptive' statements about psychopathology issue from presupposed value stances that conceal their own deeper sources, compatibilities, and incompatibilities.

It will be worth noting which values will be given priority, importance, and address in discussion and formulation of future psychiatric nosology. Our hope is that if each iteration (or revision)

of nosologic classification improves psychiatric research and practice, and consequently the health of the mentally ill, then the eventual success of nosology as *logos* – a rational study – to accurately reflect the complex reality of psychiatric illness and the soundness of its care remains promising.

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Part II

Internalizing Disorders: Neurodevelopmental Disorders

Intellectual Disabilities and Global Developmental Delay

2

Elana Pinchefsky and Michael Shevell

Overview of the History of Intellectual Disability and Global Developmental Delay in the DSMs

The diagnosis of intellectual disability (ID)/intellectual developmental disorder (IDD) and global developmental delay has evolved over time to the current definition that extends well beyond the original concepts, which were largely based on a statistical conceptualization of a general subaverage intellectual functioning, as measured by the intelligence quotient (IQ).

In the *Diagnostic and Statistical Manual of Mental Disorders*, 1st ed. (*DSM-I*; APA, 1952), intellectual disability (ID) was first termed "mental deficiency" (familial/hereditary or idiopathic) or "chronic brain syndrome with mental deficiency" if secondary. It was further divided by severity based on documented IQ into mild (IQ 70–85), moderate (IQ 50–70), and severe (IQ below 50) (APA, 1952).

The terminology changed to "mental retardation" in the *Diagnostic and Statistical Manual of Mental Disorders*, 2nd ed. (*DSM-II*; APA, 1968). Mental retardation was defined as subnormal general intellectual functioning originating during the developmental period and was associated with either deficient learning and social adjustment or maturation or both. The diagnostic classification was further subdivided in relationship to IQ as follows: borderline (IQ 68–85), mild (IQ 52–67), moderate (IQ 36–51), severe (IQ 20–35), and profound (IQ under 20) (APA, 1968). The ceiling for the diagnosis was set at one standard deviation below the mean (IQ 85), which included the lower 16% of the population and was later perceived as too broad (Greenspan & Woods, 2014). Mental retardation was then also subdivided if the associated condition or etiology was known, for example, following infection and intoxication (e.g., congenital rubella), trauma or physical agent (e.g., birth asphyxia), disorders of metabolism, etc. (APA, 1968).

The multiaxial system was introduced in the *Diagnostic and Statistical Manual of Mental Disorders*, 3rd ed. (*DSM-III*; APA, 1980). Explicit diagnostic criteria for mental retardation were introduced and included: (A) significantly subaverage general intellectual functioning (an IQ of 70 or below); (B) concurrent deficits or impairments in adaptive behavior, taking the patient's age into consideration; and (C) onset before the age of 18. The addition of adaptive behavior was intended to better reflect the social characteristics of the disability and reduce a strict reliance on IQ scores. Adaptive function was

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conventionalized to reflect how effectively an individual meets the standards of personal independence and social responsibility expected for his or her age and cultural group. Borderline intellectual functioning was no longer included in the DSM-III, since the majority of patients with borderline intellectual functioning were found not to have a significant impairment in adaptive behavior, and this diagnosis was thus changed to borderline intellectual functioning (included as a condition not attributable to a mental disorder). Consequently, there were then four degrees of severity of mental retardation: mild (IQ 50–70), moderate (IQ 35–49), severe (IQ 20–34), and profound (IQ below 20). If there was a strong presumption of mental retardation, but the patient was untestable by standard intelligence tests, they were classified as unspecified mental retardation. Etiologic factors, if known, would have been coded on axis III. DSM-III also contained outdated and now quite offensive terminology such as "idiocy," "moron," and "imbecile" (APA, 1980).

The lower maximum cutoff for the IQ score decreased false-positives, but also led to an under-identification as clinicians often used the IQ test as a screening device, and if the full-scale score was greater than 70–75, they would either decline to assess adaptive functioning or discount it when concluding a person had intellectual disability, even if there were very severe deficits observed in adaptive functioning (Greenspan & Woods, 2014).

In *Diagnostic and Statistical Manual of Mental Disorders*, 3rd ed. revised (*DSM-III-R*; APA, 1987), the category developmental disorder not otherwise specified (NOS) was introduced for disorders in development that did not meet the criteria for either "mental retardation" or a pervasive or a specific developmental disorder. Also the Axis I-Axis II distinction was better clarified and mental retardation, specific developmental disorders, and pervasive developmental disorders were classified as Axis II under Developmental Disorders (APA, 1987).

The diagnostic criteria were similar in the *Diagnostic and Statistical Manual of Mental Disorders*, 4th ed. (*DSM-IV*; APA, 1994), but were modified to be more compatible with the American Association of Mental Retardation (AAMR) consensus definition. Deficits in adaptive functioning were further defined as having to occur in at least two of the following eleven areas: communication, self-care, home living, social/interpersonal skills, use of community resources, self-direction, functional academic skills, work, leisure, health, and safety. Also, in DSM-IV, pervasive developmental disorders, learning disorders, motor skills disorder, and communication disorders were all modified to be coded on Axis I, whereas they were coded on Axis II in DSM-III-R. In DSM-IV, only personality disorders and mental retardation remained on Axis II (APA, 1994).

In *Diagnostic and Statistical Manual of Mental Disorders*, 4th ed. revised (*DSM-IV-R*; APA, 2000), additional information was added regarding some of the associated etiological factors and comorbid symptoms and disorders (e.g., fragile X syndrome (FXS) and attention-deficit/hyperactivity disorder (ADHD)) (APA, 2000).

The definition of mental retardation in the DSM-III through to the publication of DSM-IV-R was based on the classification of the American Association on Intellectual and Developmental Disabilities (AAIDD) (formerly American Association on Mental Retardation (AAMR) and before that the American Association on Mental Deficiency (AAMD)). The classification was based on a "disability" (numbers-based) approach, which changed to a more clinically relevant, neuroscience-grounded and medicine-supported more holistic "disorder" approach in the DSM-5 (Greenspan & Woods, 2014).

Current Diagnostic Criteria in DSM-5

Intellectual Disability [Intellectual Developmental Disorder (IDD)]

The *Diagnostic and Statistical Manual of Mental Disorders*, 5th ed. (*DSM-5*; APA, 2013a) attempts to broaden the category of intellectual disability/IDD and approach it in more clinical terms. Clinicians are encouraged to take into account the possible sources of test error, to not

just rely on a full-scale IQ score and rather to rely more on neuropsychological profiles of cognitive impairments, and an approach that involves assessing the adaptive functioning of patients with greater emphasis on behaviors (e.g., risk-unawareness) which reflect the underlying cognitive aspects of the disorder (Greenspan & Woods, 2014) (Table 2.1).

Table 2.1 DSM-5 diagnostic criteria for intellectual disability (intellectual developmental disorder) (American Psychiatric Association, 2013a)

Intellectual disability (intellectual developmental disorder) is a disorder with onset during the developmental period that includes both intellectual and adaptive functioning deficits in conceptual, social, and practical domains. The following three criteria must be met:

A. Deficits in intellectual functions, such as reasoning, problem solving, planning, abstract thinking, judgment, academic learning, and learning from experience, confirmed by both clinical assessment and individualized, standardized intelligence testing

B. Deficits in adaptive functioning that result in failure to meet developmental and sociocultural standards for personal independence and social responsibility. Without ongoing support, the adaptive deficits limit functioning in one or more activities of daily life, such as communication, social participation, and independent living, across multiple environments, such as home, school, work, and community

C. Onset of intellectual and adaptive deficits during the developmental period

Specify current severity:

317 (F70) Mild

318.0 (F71) Moderate

318.1 (F72) Severe

318.2 (F73) Profound

Note: The diagnostic term *intellectual disability* is the equivalent term for the ICD-11 diagnosis of *intellectual developmental disorders.* Although the term *intellectual disability* is used throughout this manual, both terms are used in the title to clarify relationships with other classification systems. Moreover, a federal statute in the United States (Public law 111–256, Rosa's law) replaces the term *mental retardation* with *intellectual disability,* and research journals use the term *intellectual disability.* Thus, *intellectual disability* is the term in common use by medical, educational, and other professions and by the lay public and advocacy groups

The term mental retardation used from DSM-II until DSM-IV-R was replaced with intellectual disability [intellectual developmental disorder (IDD)] in the *DSM-5*, as this was the term being used predominantly for the prior two decades by medical professionals, educators, and other professionals and by the lay public and advocacy groups. Furthermore, a federal statute in the United States (Public Law 111-256, Rosa's Law) replaced the term mental retardation with intellectual disability (APA, 2013a). The term intellectual developmental disorder was placed in parentheses to reflect the World Health Organization's classification system, which lists "disorders" in the International Classification of Diseases (ICD; ICD-11 to be released in 2018) and bases all "disabilities" on the International Classification of Functioning, Disability, and Health (ICF) (APA, 2013b). The name change to IDD highlights the move away from a statistical numbers-based disability emphasis for diagnosis and classification (Greenspan & Woods, 2014).

Diagnostic criteria presently utilize the need for an assessment of both cognitive capacity (IQ) and adaptive functioning. The essential features of intellectual disability (intellectual developmental disorder) are (A) deficits in general intellectual abilities; (B) impairment in everyday adaptive functioning, in comparison to an individual's age-, gender-, and socioculturally matched peers; and (C) onset that is during the developmental period. Criterion C no longer has a specified age ceiling for when the onset of actual observed impairments must be manifested (set at 18 years in the previous DSM manuals). In DSM-5 the various levels of severity of ID are determined by adaptive functioning rather than actual IQ score, since adaptive functioning is what determines the level of support an individual requires (APA, 2013a). (See Table 2.2 on severity levels for intellectual disability).

DSM-5 has moved to a nonaxial documentation; thus specifiers may be used to enhance the clinical description for the neurodevelopmental disorders, such as the age of onset or severity ratings, and may include "associated with a known medical or genetic condition or environmental factor" (e.g., fragile X syndrome).

Intellectual functioning is usually measured with individually administered and psychometrically valid, comprehensive, culturally appropriate, and psychometrically sound tests of intelligence. Individuals with intellectual disability typically have scores greater than two standard deviations or more below the population mean (IQ below 70, the second percentile), including a margin for measurement error (generally ±5 points). On tests with a standard deviation of 15 and a mean of 100, this reflects a score of between 65–75 on a test (70 ± 5) (APA, 2013a). Consequently the ceiling for intellectual disability can essentially be an IQ of 75 (the fifth percentile) (Greenspan & Woods, 2014). The individual cognitive profiles based on neuropsychological testing can identify areas of relative strengths and weaknesses, which is important for effective and accurate academic and vocational planning (APA, 2013a). There are currently no screening tools that are sufficiently valid or reliable enough to identify children with a probable intellectual disability (McKenzie & Megson, 2012).

There are many standardized measures for the clinician to choose between, with one of the most commonly used IQ tests being the Wechsler Intelligence Scale for Children, fourth edition (WISC-IV). The WISC-IV offers (1) a full-scale IQ (FSIQ), which involves scores from four domains, including verbal comprehension, perceptual reasoning, processing speed and working memory; and (2) a General Ability Index (GAI), which is calculated using only the verbal comprehension and perceptual reasoning scores (Wechsler, 2003a). A study found the GAI and FSIQ to be similarly predictive of overall adaptive functioning; however, the GAI resulted in fewer children meeting criteria for intellectual disability. The individuals that "fell out" of a diagnosis with the GAI had significantly higher adaptive functioning scores on average but still fell within the impaired range. Consequently the FSIQ was recommended for diagnostic decision-making (Koriakin et al., 2013). Clinical judgment should be used to interpret test results as various social, cultural and linguistic contexts should be considered, as well as the effect that co-occurring communication, language, motor, and/or sensory impairments may have on tests scores (Yin Foo, Guppy, & Johnston, 2013). In order to correct for out-of-date norms, it is now standard practice to lower an IQ score by 0.3 points for every year of "norm obsolescence". This is referred to eponymously as the "Flynn effect", which is essentially that individuals of today receive higher IQ scores than individuals of the past; thus obsolete norms inflate IQ scores since they measure performance against scores of test takers from the past, as opposed to higher scores from the present (Young, 2012). The DSM-5 also has a provision that highly discrepant individual subtest scores may make an overall IQ score invalid (APA, 2013a); thus something other than full-scale IQ should be used when there is excessive scatter among subscales (Greenspan & Woods, 2014).

There is now a statement in the DSM-5 that "individual cognitive profiles based on neuropsychological testing are more useful for understanding intellectual abilities than a single IQ score" (APA, 2013a). Accordingly, individuals who straddle the 70–75 IQ ceiling with severe adaptive deficits and cognitive impairments and were previously not getting the benefits of the "intellectual disability label" for eligibility of support services may now be diagnosed with ID/IDD due to the provisions added in the DSM-5 (e.g., supplementing neuropsychological measures with IQ in some circumstances, considering a diagnosis of ID/IDD when adaptive functioning is very low even if the IQ is above typical ceilings, and not relying on full-scale IQ when there is considerable scatter) (APA, 2013a; Greenspan & Woods, 2014).

Criterion B of DSM-5 requires at least one domain of adaptive functioning (conceptual, social or practical) to be sufficiently impaired that ongoing support is required for the person to perform adequately in one or more life settings (school, work, home or in the community) (APA, 2013a). Adaptive functioning refers to how well a person meets community standards of personal independence and social responsibility, in comparison to others of similar age and sociocultural background (Tassé et al. 2012). Adaptive functioning involves adaptive reasoning in three domains:

Table 2.2 Severity levels for intellectual disability (intellectual developmental disorder) (American Psychiatric Association, 2013a)

Severity level	Conceptual domain	Social domain	Practical domain
Mild	*Preschool children*:: • There may be no obvious conceptual differences *School-age children and adults*: • There are difficulties in learning academic skills involving reading, writing, arithmetic, time, or money, with support needed in one or more areas to meet age-related expectations *Adulthood*: • Abstract thinking, executive function (i.e., planning, strategizing, priority setting, and cognitive flexibility), and short-term memory, as well as functional use of academic skills (e.g., reading, money management), are impaired. There is a somewhat concrete approach to problems and solutions compared with age-mates	Compared with typically developing age-mates, the individual is immature in social interactions • There may be difficulty in accurately perceiving peers' social cues • Communication, conversation, and language are more concrete or immature than expected for age • There may be difficulties regulating emotion and behavior in age-appropriate fashion • These difficulties are noticed by peers in social situations There is limited understanding of risk in social situations; social judgment is immature for age, and the person is at risk of being manipulated by others (gullibility)	May function age-appropriately in personal care Need some support with complex daily living tasks in comparison to peers *Adulthood*: • Supports typically involve grocery shopping, transportation, home and child-care organizing, nutritious food preparation, and banking and money management • Recreational skills resemble those of age-mates, although judgment related to well-being and organization around recreation requires support • Competitive employment is often seen in jobs that do not emphasize conceptual skills • Generally need support to make health-care and legal decisions and to learn to perform a skilled vocation competently • Support is typically needed to raise a family
Moderate	All through development, the individual's conceptual skills lag markedly behind those of peers *Preschool children*: • Language and pre-academic skills develop slowly *School-age children*: • Progress in reading, writing, mathematics, and understanding of time and money occurs slowly and is markedly limited compared with that of peers *Adulthood*: • Academic skill development is typically at an elementary level • Support is required for all use of academic skills in work and personal life • Ongoing assistance on a daily basis is needed to complete conceptual tasks of day-to-day life, and others may take over these responsibilities fully for the individual	The individual shows marked differences from peers in social and communicative behavior across development • Spoken language is typically a primary tool for social communication but is much less complex than that of peers • Capacity for relationships is evident in ties to family and friends, and the individual may have successful friendships across life and sometimes romantic relations in adulthood • Individuals may not perceive or interpret social cues accurately • Social judgment and decision-making abilities are limited, and caretakers must assist the person with life decisions • Friendships with typically developing peers are often affected by communication or social limitations • Significant social and communicative support is needed in work settings for success	*Adulthood*: • Can care for personal needs involving eating, dressing, elimination, and hygiene, although an extended period of teaching and time is needed for the individual to become independent in these areas, and reminders may be needed • Participation in all household tasks can be achieved, although an extended period of teaching is needed, and ongoing supports will typically occur for adult-level performance • Independent employment in jobs that require limited conceptual and communication skills can be achieved, but considerable support from co-workers, supervisors, and others is needed to manage social expectations, job complexities, and ancillary responsibilities such as scheduling, transportation, health benefits, and money management • A variety of recreational skills can be developed. These typically require additional supports and learning opportunities over an extended period of time Maladaptive behavior is present in a significant minority and causes social problems

(continued)

Table 2.2 (continued)

Severity level	Conceptual domain	Social domain	Practical domain
Severe	Attainment of conceptual skills is limited • Generally has little understanding of written language or of concepts involving numbers, quantity, time and money • Caretakers provide extensive supports for problem solving throughout life	Spoken language: • Quite limited in terms of vocabulary and grammar • Speech may be single words or phrases and may be supplemented through augmentative means • Speech and communication are focused on the here and now within everyday events • Language is used for social communication more than for explication • Understand simple speech and gestural communication Relationships with family members and familiar others are a source of pleasure and help	The individual requires support for all activities of daily living, including meals, dressing, bathing, and elimination • The individual requires supervision at all times The individual cannot make responsible decisions regarding well-being of self or others *Adulthood*: • Participation in tasks at home, recreation, and work requires ongoing support and assistance • Skill acquisition in all domains involves long-term teaching and ongoing support Maladaptive behavior, including self-injury, is present in a significant minority
Profound	Conceptual skills generally involve the physical world rather than symbolic processes The individual may use objects in a goal-directed fashion for self-care, work, and recreation Certain visuospatial skills, such as matching and sorting based on physical characteristics, may be acquired. However, co-occurring motor and sensory impairments may prevent functional use of objects	The individual has very limited understanding of symbolic communication in speech or gesture • May understand some simple instructions or gestures • Expresses his or her own desires and emotions largely through nonverbal, nonsymbolic communication Social: • Enjoys relationships with well-known family members, caretakers, and familiar others and initiates and responds to social interactions through gestural and emotional cues • Co-occurring sensory and physical impairments may prevent many social activities	The individual is dependent on others for all aspects of daily physical care, health, and safety, although he or she may be able to participate in some of these activities as well • Individuals without severe physical impairments may assist with some daily work tasks at home, like carrying dishes to the table • Simple actions with objects may be the basis of participation in some vocational activities with high levels of ongoing support Recreational activities: • May involve, for example, enjoyment in listening to music, watching movies, going out for walks, or participating in water activities, all with the support of others • Co-occurring physical and sensory impairments are frequent barriers to participation (beyond watching) in home, recreational, and vocational activities Maladaptive behavior is present in a significant minority

1. *Conceptual (academic) domain*: competence in memory, language, reading, writing, math reasoning, acquisition of practical knowledge, problem solving, and judgment in novel situations.
2. *Social domain*: awareness of others' thoughts, feelings, and experiences, empathy, interpersonal communication skills, friendship abilities and social judgment.
3. *Practical domain:* learning and self-management across life settings, including personal care, job responsibilities, money management, recreation, self-management of behavior, and school and work task organization (APA, 2013a).

Adaptive functioning is assessed using both clinical evaluation and individualized, culturally appropriate, psychometrically sound measures. Standardized measures are used by gathering evidence from knowledgeable informants (e.g., parent or other family member, teacher, counselor, care provider) and from the individual when possible. Additional sources of information include educational, developmental, medical and mental health evaluations. Several factors may influence adaptive functioning, including intellectual capacity, education, motivation, socialization, personality features, vocational opportunity, cultural experience and coexisting general medical conditions or mental disorders (APA, 2013a).

Several age-appropriate scales have been designed to help quantify adaptive functioning or behavior. Scores for certain individual domains are not included in some of these instruments, and individual domain scores may vary considerably in reliability (APA, 2000). When standardized testing is difficult or impossible, because of a variety of factors (e.g., sensory impairment, severe problem behavior, language barrier), the individual may be diagnosed with an unspecified intellectual disability (APA, 2013a).

Consequently, relying just on the full-scale IQ is now outdated, and using cross-battery profile methods of cognitive strengths and weaknesses across many tests (including neuropsychological measures) is currently seen as much more diagnostically substantive and much more likely to capture the complex individuality of the persons being evaluated (APA, 2013a; Greenspan & Woods, 2014; McGrew & Flanagan, 1997).

Global Developmental Delay

The term global developmental delay (GDD) was only introduced in the DSM-5 but had emerged prior as an entity to describe a disturbance across a variety of developmental domains (Batshaw & Shapiro, 1997; Fenichel, 2001; Kinsbourne & Graf, 2001; Majnemer & Shevell, 1995; Shevell, Majnemer, Rosenbaum, & Abrabamowicz, 2000; Simeonsson & Simeonsson, 2001). GDD is diagnosed when a child fails to meet expected developmental milestones in several domains of function. The diagnosis is reserved for children under the age of 5 years, when they are unable to reliably do systematic assessments of intellectual functioning or are too young to undergo standardized testing (APA, 2013a).

The American Academy of Neurology (AAN) practice parameter published in 2003 defines GDD operationally as a significant delay in two or more developmental domains: (1) gross/fine motor, (2) speech/language, (3) cognition, (4) social/personal, and (5) activities of daily living. Significant delay is defined as performance two standard deviations or more below the mean on age-appropriate, standardized norm-referenced testing. Typically, delays in development are noted across all developmental domains (Shevell et al., 2003). Importantly, an infant or child with a global developmental delay (e.g., due to a neuromuscular disorder or perinatal asphyxia) will not necessarily have an intellectual disability when they are old enough to measure cognitive level. However, both ID/IDD and GDD share complementary features, and both represent neurodevelopmental disabilities that share the essential feature of a predominant disturbance in the acquisition of cognitive, motor, language or social skills. Thus, a common approach to their evaluation and understanding their etiology can be used (Sherr & Shevell, 2012; Shevell, 2008).

Etiology

Many disorders have no specific therapeutic interventions; however, the determination of the etiology of ID has been shown to help guide follow-up management and referral recommendations. Several etiologies may have specific implications for treatment, prognosis, identification and medical management of associated conditions, assessment of recurrence risk, and preventing recurrence through screening for carriers and prenatal testing. Accurate etiologic diagnosis may also empower the affected family in planning for their child, improving the understanding of treatment and prognosis, limiting further unnecessary testing which may be costly and invasive, as well as improved access to available services (Coulter et al., 2011; Majnemer & Shevell, 1995; Makela, Birch, Friedman, & Marra, 2009; Michelson et al., 2011; Saam, Gudgeon, Aston, & Brothman, 2008; Shevell, 2008, 2009; Yeargin-Allsopp, Murphy, Cordero, Decoufle, & Hollowell, 1997).

The most common preventable cause of ID in the Western world, particularly in the United States, is fetal alcohol syndrome (FAS) (May & Gossage, 2001). FAS consists of characteristic facial dysmorphic features, growth deficiency, and central nervous system dysfunction. Fetal alcohol spectrum disorder (FASD) describes the spectrum of effects in offspring with prenatal alcohol exposure due to maternal alcohol consumption who demonstrate some, but not all of the features of FAS, but they may have alcohol-induced cognitive impairments (Spohr, Willms, & Steinhausen, 2007).

In industrialized countries, preventive measures have had significant impact for specific causes of ID. These include prevention of congenital hypothyroidism though the introduction of iodized table salt, systematic newborn screening for phenylketonuria and a variety of other treatable disorders, antibiotics for congenital syphilis, and the near eradication of congenital rubella syndrome through community-wide vaccination efforts (Silverman, 2009). It is estimated that these medical interventions have prevented approximately 16% of cases of ID that would have occurred without specific medical intervention (Brosco, Sanders, Dowling, & Guez, 2013). Congenital hypothyroidism still remains among the most common preventable causes of intellectual impairment in many underdeveloped regions of the world (Jain, Agarwal, Deorari, & Paul, 2008; Tikaria et al., 2010). Interestingly, there is a strong genetic component to the link between iodine deficiency and ID because specific alleles for the deiodinase type II gene and the ApoE4 allele confer a significantly greater risk of ID when the pregnant mother is iodine-deficient (Guo et al., 2004; Wang et al., 2000). Also, only a minority of the children born to women that are defined as heavy consumers of alcohol have FAS, and twin studies suggest that at least part of this susceptibility to alcohol teratogenesis is genetically determined (Streissguth & Dehaene, 1993).

The prevalence of ID is inversely correlated with socioeconomic standards, both within and between countries (Drews, Yeargin-Allsopp, Decoufle, & Murphy, 1995; Gustavson, 2005). Other specific risk factors for ID include low birth weight, prematurity, and diverse prenatal and/or perinatal complications (Brosco et al., 2013; Hack et al., 2002; Yager & Gressens, 2009). Low birth weight is associated with approximately 10–15% of the total prevalence of ID (Brosco et al., 2013). In underdeveloped regions of the world, the prevalence of ID is two- to threefold higher than in high-income countries (Durkin, 2002; Gustavson, 2005). This has been attributed to a variety of factors, including malnutrition, parental illiteracy, limited intellectual stimulation, and poor health care, but also to high rates of parental consanguinity (Christianson et al., 2002; Jazayeri, Saberi, & Soleymanzadeh, 2010; Sachdeva et al., 2010).

A study by Jauhari et al. in Northern India found that 75% of the etiologies identified were perinatal (predominantly asphyxia or infection) or postnatal (exclusively central nervous system infection) in origin, and these accounted for 40% of all cases of intellectual disability or global developmental delay evaluated (Jaurari et al., 2011). Whereas in Western series, the majority are due to prenatal (genetic) causes. Srour, Mazer, and Shevell (2006) found in a community-derived

ambulatory clinic population with GDD in Canada that the most common etiologies were genetic syndromes/chromosomal abnormalities (24%), intrapartum asphyxia (22%), cerebral dysgenesis (16%), and psychosocial deprivation (11%) (Srour et al., 2006).

Genetic Causes

Significant progress has been made in the elucidation of the genetic factors underlying ID. Mutations in more than 450 genes have been identified, yet the precise genetic cause remains unknown in the vast majority of individuals with ID (Ropers, 2010; van Bokhoven, 2011). Mild forms of ID are thought to represent the lower end of the normal IQ distribution and to result from a complex interplay of many genetic and environmental factors. Due to its genetic complexity, still little is known about the genetic factors predisposing to mild ID (Ropers, 2010). A conclusive genetic or metabolic diagnosis can be made in approximately 50–65% of patients with moderate-to-severe ID, in contrast to 20% for mild ID cases (van Bokhoven, 2011).

Down syndrome/Trisomy 21 is the most frequent genetic form of ID with a prevalence of 1/750–1/800. Its frequency has remained remarkably constant in the United States and in most parts of the world despite widespread prenatal diagnosis in older mothers (Besser, Shin, Kucik, & Correa, 2007; Centers for Disease Control and Prevention, 2006; Collins, Muggli, Riley, Palma, & Halliday, 2008; Mefford, Batshaw, & Hoffman, 2012).

Fragile X syndrome (FXS), one of the most common genetic causes of ID, is found in 2–2.5% of cognitively impaired males and in approximately 25% of males with X-linked ID (XLID) (Ropers, 2010). FXS has an estimated prevalence of 1 in 3000 males (Crawford, Acuna, & Sherman, 2001). A mutation of the FMR1 gene leads to transcriptional silencing and is mediated by expansion of a CGG trinucleotide repeat in the 5′ end of the gene (Verkerk et al., 1991). Four forms of the CGG trinucleotide repeat have been described: normal (6–40 repeats), intermediate (41–60 repeats), premutation (61–200 repeats) and full mutation (>200–230 repeats) (Crawford et al., 2001). Patients with FXS have narrow and elongated faces, large protruding ears, macroorchidism, joint hyperlaxity and up to 20% may have epilepsy (Willemsen, Oostra, Bassell, & Dictenberg, 2004). Carrier females and males with somatic mutations have various levels of intellectual impairment (Willemsen et al., 2003). Initial studies suggested that premutation carriers (55–200 repeats) were asymptomatic; however, it was later shown that premutation carriers can have primary ovarian insufficiency (if female) or fragile X-associated tremor/ataxia syndrome (FXTAS) (intention tremor and cerebellar ataxia). These FXTAS individuals may experience evidence of cognitive decline after the age of 50 years, likely due to a toxic "gain of function" of the abnormal FMR1 mRNA (Hagerman & Hagerman, 2004; Jacquemont et al., 2004).

It has been observed that moderate-to-severe ID is approximately 1.4 times more frequent in males than in females (Leonard & Wen, 2002) and that the male excess appears to be particularly significant in mild ID (McLaren & Bryson, 1987). The relative ease of detecting the familial transmission of X-chromosome mutations has facilitated efforts to uncover ID genes on the X-chromosome. Currently, it is believed that in males, X-linked recessive gene defects account for about 10–15% of ID (deBrouwer et al., 2007; Ropers & Hamel, 2005; van Bokhoven, 2011). XLID is a very heterogeneous group of disorders, subdivided into syndromic (S-XLID) and nonsyndromic forms (NS-XLID). S-XLID is associated with other, often specific clinical findings, which are key for the selective targeted identification of the underlying molecular defects. Finding the molecular causes of NS-XLID is a much greater challenge because genetically distinct subtypes are clinically indistinguishable and thus investigators cannot pool data from unrelated families (Ropers, 2010). Over 90 different genes have been implicated in XLID, and mutations in over 40 of these have been found in patients with NS-XLID (Ropers, 2010; Tarpey et al., 2009). The establishment of large cohorts of clinically well-characterized XLID families in

consortia such as EURO-MRX (deBrouwer et al., 2007) and iGOLD (Tarpey et al., 2009) have helped to accelerate the successful elucidation of NS-XLID genes.

One of the first genes implicated in NS-XLID was *OPHN1*, a Rho-GTPase-activating protein (Billuart et al., 1998); however, it was later found to also be associated with a syndrome including ataxia, epilepsy and cerebellar hypoplasia (Bergmann et al., 2003). In the EURO-MRX cohort, OPHN1 mutations had a prevalence of 5% (deBrouwer et al., 2007). Moreover, mutations in genes regulating the activity of small Rho and Ras GTPases were among the first identified in patients with syndromic and nonsyndromic forms of XLID (Billuart et al., 1998; Kutsche et al., 2000). These genes are known to play an important role in neuronal development and synapse formation and function. Genes thus far implicated in XLID play a role in many other processes, including cell adhesion, transcription regulation, posttranslational modification, signal transduction, ubiquitination, actin cytoskeleton modification and sodium transport (Ropers, 2010; Tarpey et al., 2009). Testing of XLID genes has a yield of 42% in males from definitely X-linked families and 17% in males from possibly X-linked families (deBrouwer et al., 2007; Michelson et al., 2011).

Cytogenetically detectable and submicroscopic chromosomal rearrangements account for approximately 25% of all cases of ID (Ropers, 2010). Recurrent microdeletions detectable by conventional karyotyping or by targeted fluorescence in situ hybridization (FISH) analysis are found in a number of conditions with recognizable clinical features, including Prader-Willi and Angelman (15q11.2-q13), Williams (7q11.23), Smith-Magenis (17p11.2), and DiGeorge (22q11.2), most of which have now been explained by haploinsufficiency of specific genes (Goldenberg & Saugier-Veber, 2010; Lossie et al., 2001; Sahoo et al., 2008). Deletions and duplications too small to be detectable by conventional karyotyping are now routinely identified by array comparative genomic hybridization (aCGH).

The number of genes linked to ID has dramatically increased in recent years with the introduction of aCGH and whole exome sequencing (WES). aCGH has become the method of choice for detecting deletions or duplications in the clinical setting, or more generally, copy number variants (CNVs), which have been found throughout the human genome (Ropers, 2010). Currently a variety of commercial oligonucleotide-based single nucleotide polymorphism (SNP) microarrays are commonly used with increasing genomic coverage of nearly 3 million small synthetic oligonucleotide probes per array (van Bokhoven, 2011).

aCGH studies have revealed an abundance of apparently pathogenic CNVs in 7.8–20% of patients with idiopathic ID and normal karyotype, depending on the preselection of tested patients (de Vries et al., 2005; Hochstenbach et al., 2009; Jaillard et al., 2009; Koolen et al., 2009; Manning & Hudgins, 2007; Michelson, 2011; Miller, 2010; Sagoo et al., 2009). CNVs are the fundamental cause of many specific forms of ID and varied CNVs (e.g., at 1q21.1, 15q11.2, 15q13.3, 16p11.2, 16p13.11, and 22q11.2) are also major predisposing factors for a wide spectrum of neuropsychiatric disorders, including autism spectrum disorder (ASD), schizophrenia, and possibly bipolar affective psychosis. Strict regulation of gene dose in neurodevelopment may be a critical brain mechanism in fostering a susceptibility to disease (Morrow, 2010; Ropers, 2010). CNVs are also more common in patients with both ID and generalized epilepsy than ID or generalized epilepsy alone (Mullen et al., 2013). Sometimes the clinical features of recurrent CNVs are consistent enough for the definition of a new ID syndrome, for example, the chromosome 17q21.31 deletion syndrome (Koolen et al., 2006), whereas other microdeletions and duplications are associated with multiple phenotypes of varying severity (Girirajan et al., 2012). Also, a few cases reveal CNVs which are a single dosage-sensitive gene as the cause of the ID phenotype, such as *CHD7* deletions in CHARGE syndrome (Vissers et al., 2004), and deletions of *EHMT1* in Kleefstra syndrome (Kleefstra et al., 2005).

WES is a high-throughput technique that involves sequencing all the protein-coding regions (exons) in the human genome (Srivastana et al., 2014). There are approximately 20,000 protein-coding genes (~2% of the total sequence). The remaining 98% of noncoding genome consists of mobile elements and regulatory regions and elements (promotors, noncoding RNAs, introns, etc.), the majority of which still have a currently unknown function. Whole exome sequencing of ID patients has revealed more than 60 novel genes implicated in ARID (Caliskan et al., 2011; Najmabadi et al., 2011) or nonsyndromic sporadic ID with de novo dominant single-gene mutations (De Ligt et al., 2012; Rauch et al., 2012; Vissers et al., 2010). Rauch et al. used WES in 51 patients with severe nonsyndromic ID and found that 16 (31%) had mutations in known ID genes (Rauch et al., 2012). A study of 100 individuals with unexplained ID (IQ < 50) found the diagnostic yield of WES was 16% (13% had a molecular diagnosis in known intellectual disability genes, and de novo mutations in three candidate genes were identified) (De Ligt et al., 2012).

Additionally, a significant overlap in ID and ASD genes has been found, and many of the genes are part of common pathways involved in synaptic plasticity (Kou, Betancur, Xu, Buxbaum, & Ma'ayan, 2012; van Bokhoven, 2011). This overlap was highlighted by the discovery that 20% of the novel genes identified with WES in ASD patients were targets of FMRP (fragile X protein), the most recognized genetic cause of ID (Iossifov et al., 2012). Clinically this overlap is also apparent in that 40% of ID patients have ASD and 50–85% of ASD patients have concurrent ID (Brereton, Tonge, & Einfeld, 2006; Matson & Shoemaker, 2009).

There are a number of limitations of WES, which include that WES will detect only exon and splicing site changes; it will not detect large insertions/deletions, structural chromosomal rearrangements, trinucleotide repeats, intronic alterations, epigenetic modifications, or mutations in non-exonic regulatory regions. There are also technical limitations, in that some exon sequences are not well covered by the sequencing process. Additionally, mitochondrial DNA must be sequenced via separate technology. Furthermore, the neurodevelopmental phenotype of a patient may be the result of complex interactions among different gene networks and modifying environmental factors. Finally, analysis performed at commercial WES laboratories may exclude, or not be able to interpret, certain variants particularly those genes with unknown function (Srivastana et al., 2014).

ID and Autism Affect Synaptic Morphology and Plasticity, and Epigenetic Control

Growing evidence suggests that ID and ASD share a common molecular etiology at a single-gene level (van Bokhoven, 2011). Mutations in many different types of ID and ASD genes affect synaptic morphology and plasticity, as well as epigenetic control of gene expression. Consequently, molecular networks involving ID genes can be grouped in the following broad functions:

1. Presynaptic pathways (neurotransmitter release)
2. Postsynaptic protein complexes (receptor activity)
3. Cytoskeleton dynamics
4. Intracellular signal transduction pathways [e.g., Ras signaling pathway (neuro-cardio-facio-cutaneous conditions)], and the PI3K-mTOR pathway (e.g., FXS, tuberous sclerosis, Cowden disease, Rett syndrome)
5. Transcription regulation
6. Epigenetic modulation of the chromatin structure (van Bokhoven, 2011)

Structural modifications of the synapse are an important focus in ID. Synapses occur mostly between axons and dentrites, at substructures called spines, which are highly dynamic protrusions composed mostly of actin but also connected to dendritic microtubules. Changes in spine shape (synaptic plasticity) have been

correlated with learning in long-term potentiation (Muller, Toni, & Buchs, 2000). The mechanisms involved in these synaptic changes are beginning to be uncovered and appear to include the disassembly of adhesion molecules that span the synaptic cleft, the breakdown of actin filaments in the postsynaptic density (PSD), and the trafficking of 2-amino-3-hydroxy-5-methyl-4-isoxazole propionic-acid (AMPA) glutamate binding (excitatory) receptors. Knowledge of these processes has significantly advanced our understanding of the function of the normal and disrupted synaptic functions of ID-associated proteins (van Bokhoven, 2011).

Multiple presynaptic proteins have been involved in ID such as genes involved in neurotransmitter vesicles release (which have also been frequently associated with epilepsy), such as STXBP1 (ID and Ohtahara seizure syndrome) and synapsin I (van Bokhoven, 2011).

Postsynaptic protein networks and pathways have also been shown to be key in cognition and ID. Systematic mutation screening in a cohort of 94 unrelated patients with idiopathic ID revealed three de novo heterozygous truncating mutations in the *SYNGAP1* gene. *SYNGAP1* is expressed in the synapse and mediates synaptic plasticity by controlling the insertion of AMPA receptors into the synaptic membrane. SYNGAP1 is a cause for autosomal dominant nonsyndromic ID in approximately 3–5% of cases (Hamdan et al., 2009, 2011).

Several proteins in the GTPase family regulate the actin and microtubule cytoskeleton. Oligophrenin (OPHN1, discussed above) is one of several proteins of the RhoGTPase family. OPHN1 also has a role in synapse formation and function (presynaptic) and controls synapse maturation and plasticity by stabilizing postsynaptic AMPA receptors (Nadif Kasri, Nakano-Kobayashi, Malinow, Li, & Van Aelst, 2009; van Bokhoven, 2011). Other RhoGTPase proteins involved in actin cytoskeleton dynamics that are associated with ID include FGD1, MEGAP, ARHGEF9/collybistin, ARHGEF6, PAK3, and LIMK1 (Arber et al., 1998; Ba, van der Raadt, & Kasri, 2013; Endris et al., 2002; van Bokhoven, 2011).

Control of gene expression at the epigenetic level can be affected by DNA methylation, which is deficient in Rett syndrome. Rett syndrome is believed to be one of the leading causes of severe intellectual disability in females and is caused by mutations in the X-linked methyl-CpG binding MeCP2 gene. Patients with classic Rett syndrome appear to develop normally until 6–12 months of age, then gradually lose speech and purposeful hand use, and develop an abnormal deceleration of head growth that leads to secondary microcephaly. Seizures, autistic-like behavior, ataxia, intermittent hyperventilation and stereotypic hand movements (e.g., hand-wringing) occur in most patients. MECP2 mutations have subsequently been identified in patients with a variety of clinical syndromes ranging from mild learning disability in females to severe intellectual disability, seizures, ataxia, and sometimes severe neonatal encephalopathy in males (Shahbazian & Zoghbi, 2001).

Angelman syndrome, FXS, Coffin-Lowry syndrome and Rubinstein-Taybi syndrome also have a known epigenetic origin (Urdinguio, Sanchez-Mut, & Esteller, 2009; van Bokhoven, 2011). In Rubinstein-Taybi syndrome, gene expression is affected by a mutation in the gene CREB binding protein (CREBBP), which is a histone acetyltransferase. Additionally, various members of signaling complexes that functionally interact are found to have genes mutated in patients with ID. This is demonstrated with proteins known to form complexes with MECP2 such as ATRX, DNMT3b, CBP, RSK, CDKL5 and are encoded by genes implicated in ID (Kramer & van Bokhoven, 2009; Urdinguio et al., 2009). Gene expression can also be altered by affecting transcription factors, for example, mutations in certain zinc finger proteins have been shown to cause NSID (Ramaswamy, Castillo, & Bolduc, 2010).

Metabolic Disorders

In Caucasians, inborn errors of metabolism (IEM) account for approximately 1% of NSID (van Karnebeek, Jansweijer, Leenders, Offringa,

& Hennekam, 2005). Individually these disorders are quite rare, but collectively they are not. IEM are important to identify since specific treatment that modifies outcome is often available. Examples of IEMs associated with ID include phenylketonuria, congenital disorders of glycosylation (CDGs), creatine transporter disorders, Smith-Lemli-Opitz syndrome, peroxisomal disorders, and mitochondrial disorders, among others (Michelson et al., 2011). Most IEMs are autosomal recessive disorders, but some of the more common ones are X-linked recessive (e.g., deficiency of the creatine transporter SLC6A8) (Salomons et al., 2001). A recent systematic review of the literature identified 81 treatable IEM with ID as a major clinical finding. The majority (60%) of these conditions can be detected through a panel of widely available screening tests on blood and urine, but the remaining conditions require a "single test per single disease" approach, including single metabolite or primary molecular analysis. Each individual disorder can have a variety of different clinical presentations. A web application (www.treatable-id.org) can facilitate the recognition of treatable etiologies of ID and maximize the efficiency of diagnostic workup (van Karnebeek, Houben, Lafek, Giannasi, & Stockler, 2012).

Prevalence

The prevalence of GDD in the pediatric population is not precisely known; however, estimates of affected children range between 1% and 3% (Yeargin-Allsopp et al., 1997). ID may affect 1.5–5% of the populations in Western countries (Leonard & Wen, 2002).

Challenges in the epidemiology of intellectual disability relate to the population studied and how ID is defined and classified and how these definitions may change over time. There is a high degree of variation in estimates around the expected value, which is far more obvious for mild ID than for severe ID, as mild ID may be affected by frequently occurring external environmental factors (e.g., poor education, poor nutrition, environmental exposures, maternal health conditions, poverty, limited intellectual stimulation). Nonetheless, a review by Leonard and Wen (2002) found the prevalence rates for mild ID in 15 studies varied from 5 to 80 cases per 1000 people, with an average prevalence of 35 per 1000. In contrast, in 37 studies of severe ID, the prevalence varied only between 2.5 and 7 per 1000, with an average of 3.6 per 1000 (Leonard & Wen, 2002).

Larson and Lakin (2010) found that the estimated prevalence for intellectual disability in 2008 in elementary and secondary students in the United States was 0.85%. They reviewed US Department of Education's Accountability Center data between 1999 and 2008 and found that the number of students in the United States with a primary diagnosis of intellectual disability decreased by 20.4%, whereas there was a large increase in the number of students with autism (349%) and developmental delay (390.6%). Some of the difference was thought to likely be due to the increased use of the developmental delay and autism labels (Larson & Lakin, 2010).

Based on the National Health Interview Surveys of US households approximately one in six children in the United States in 2006–2008 were reported to have a developmental disability. Boys had twice the prevalence of any developmental disability, and there was nearly a twofold higher prevalence of any reported developmental disability (including ID) among children insured by Medicaid relative to those insured by private insurance. The prevalence of intellectual disability was found to be 0.67% in 2006–2008 (percent change of −1.5 compared to 1997–1999). One of the factors thought to be influencing this change was that since 1997, federal law has allowed for state and local education agencies to extend the use of the less-specific "developmental delay" category up to 9 years of age, enabling many children to not require a more specific education classification, such as intellectual disability. Some of these children may have been identified by the diagnostic label "other developmental delay," as suggested by the high and increasing prevalence for this category (24.7% change). The estimated prevalence of other developmental delay was 4.24% in 2006–2008 (Boyle et al., 2011).

In addition to gender disparity, the Metropolitan Atlanta Developmental Disabilities Study found a racial disparity in the prevalence of ID, with a higher prevalence of mild ID found in African-American children relative to the Caucasian population (Yeargin-Allsopp, Drews, Decoufle, & Murphy, 1995).

Symptom Presentation

Infants and children with ID generally do not reach developmental milestones within the expected age range for infants and children of their same age and cultural environment. "Red flags" for significant delay in development, which usually prompt referral to specialist, are summarized in Table 2.3. Depending on the etiology and severity of ID, there may be variable age and characteristic features at onset. The age at presentation is usually inversely proportional to the severity of ID. Most children with severe ID are recognized within the first 2 years with delay in most developmental domains. Most children with milder ID can be detected by 3–4 years of age with careful surveillance for delays of language, visual problem solving, and adaptive skills. However, many children who have mild-to-borderline ID may have an early development within normal limits and may not be identified till school age when they present with learning difficulties or poor attention skills that prompt formal IQ testing. However, the child with poor attention may not meet criteria for ADHD, but the child's ability to stay on task and concentrate may actually be consistent with his or her mental rather than chronologic age (Walker & Johnson, 2006).

Table 2.3 Red flags for referral for developmental delay

Age	Red Flags
2 months	Lack of fixation
4 months	Lack of steady head control Lack of visual tracking
6 months	Failure to turn to sound or voice Lack of smile
9 months	Inability to sit Lack of babbling consonant sounds Lack of reciprocal vocalizations or smiles
12 months	Failure to respond to name Lack of reciprocal gestures (e.g., showing, reaching, waving)
15 months	Lack of single words Lack of proto-declarative gestures (e.g., pointing or showing)
18 months	Inability to walk Failure to follow a simple command without a gesture Lack of simple pretend play Lack of spoken language/gesture combinations
24 months	Failure to use 10 to 25 single words
26 months	Failure to speak in two-word sentences
36 months	Failure to speak in three-word sentences Unintelligible speech
Any age	*Regression or loss of any previously acquired skills*

Gerber, Wilks, and Erdie-Lalena (2010a, 2010b), Johnson, Walker, Palomo-Gonzalez, and Curry (2006), Wilks, Gerber, and Erdie-Lalena (2010)

If there is an acquired etiology to the ID, the onset may be abrupt following an illness such as meningitis, encephalitis, or a head trauma occurring during early life. When intellectual disability results from a loss of previously acquired cognitive skills, as in severe traumatic brain injury, the diagnoses of intellectual disability and of a neurocognitive disorder may both be assigned (APA, 2013a).

Rarely parents present to their pediatrician with a primary concern of ID, usually children present with speech delay, the most common type of developmental delay. All children with a language delay should have formal audiologic evaluation, regardless of a normal newborn hearing screen. A speech and language pathologist should determine whether the child has an isolated expressive language (speech) delay or a more serious combined expressive and receptive language delay. Children with delayed expressive language but normal receptive language usually will overcome this temporary lag in expression and will not have eventual ID (Aylward, 2004; Walker & Johnson, 2006). Sometimes parents may become concerned when their child is unable to eat, dress, or toilet independently, especially when compared to other siblings (Walker & Johnson, 2006). Patients with severe ID may present with stereo-

typic motor movements (similar to those seen in ASD) and self-injurious behaviors (e.g., head banging, self-biting) (Walker & Johnson, 2006). One study found that children with ID have a specific profile of weaknesses in executive functioning compared to mental age-matched controls with difficulties particularly in inhibition of salient but incorrect responses, planning, and nonverbal executive-loaded working memory (Danielsson, Henry, Messer, & Ronnberg, 2012).

Features associated with ID may include difficulties with social adjustment in social relationships, such as social information processing, social interactions (including socio-emotional regulation), emotional expression and regulation, assessment of risk, or motivation in school or work environments (Baurain, Nader-Grosbois, & Dionne, 2013; Cook & Oliver, 2011; Fiasse & Nader-Grosbois, 2012). Lack of communication skills may lead to disruptive and aggressive behaviors as a substitute for communicative language (Hubert-Williams & Hastings, 2008).

Individuals with ID are much less able to recognize and avoid risks. Gullibility (induced "social foolishness" or naiveté in social situations and a tendency to being easily misled by others) is also often a feature. Gullibility and lack of an overt awareness of risk may result in exploitation by others and possible victimization, fraud, unintentional criminal involvement (due to greater susceptibility of being manipulated into participating in crimes they otherwise would have little incentive to commit), false confessions (due to greater susceptibility to be manipulated by interrogators into confessing to crimes they may not have committed), and risk for physical and sexual abuse. These associated features can have substantive implications in criminal cases (Greenspan, 2008; Greenspan, Switzky, & Woods, 2011).

The pediatric patient with GDD or ID can be vulnerable with respect to ethical issues due to the absence of capacity for competence (defined as "the capacity to understand the context of the decision, the choices available, the likely outcomes of the varying choices, and to rationally process this information to make a decision"), as well minority age (typically less than 18 years) (Bernat, 2002). For those considered unable to provide informed consent, a responsible proxy decision-maker should be selected considering the best interests of the individual and someone who can consider the risks and benefits for intervention from the unique perspective of the affected individual. The preferred proxy is usually the child's parents/family members (Shevell, 1998).

Associated Impairments and Comorbidities

Children with ID or GDD frequently have a range of comorbid mental, neurodevelopmental, medical and physical conditions, some of which may be associated with specific etiological factors. Associated problems may be internalizing (e.g., anxiety or depression) and/or externalizing (e.g., tantrums, self-injurious behaviors, aggression, ADHD, and stereotypic mannerisms or obsessive-compulsive behaviors). The challenging behaviors may cause parental stress, affect the quality of life, and result in individual and/or familial psychological, social, and economic burdens (Oeseburg, Jansen, Dijkstra, Groothoff, & Reijneveld, 2010a; Tervo, 2012; Totsika & Hastings, 2009).

Some of these associated problems may be apparent at an early age, whereas other behaviors emerge later. The most prevalent chronic health conditions in children (0–18 years) with ID include epilepsy, cerebral palsy, anxiety, oppositional defiant disorder, Down syndrome and ASD (Oeseburg, Dijkstra, Groothoff, Reijneveld, & Jansen, 2011); whereas the most prevalent chronic health conditions in adolescents (12–18 years old) include ADHD, pervasive developmental disorder not otherwise specified (PDD-NOS), dyslexia, migraine or chronic headaches and ASD. Adolescents with more severe ID had a higher prevalence of chronic mental diseases (Oeseburg et al., 2010a).

Since many individuals with intellectual limitations may be unable to express themselves, primary caretakers are vital in identifying symptoms such as eating problems, sleep problems, irritability, mood dysregulation, or aggression.

Psychiatric Disorders

The prevalence of psychiatric conditions is about three times higher among individuals with ID than in the general population (Wallander, Dekker, & Koot, 2006). Behavioral difficulties may be far more disabling to the child and family than the cognitive or language disabilities and are thus crucial to identify and treat to improve the individual's quality of life (Marco, 2009a). The diagnosis of co-occurring challenging behaviors or mental and neurodevelopmental disorders is especially challenging in minimally verbal individuals and requires modification based on developmental and cognitive abilities and focuses on behavioral domains and inferential diagnosis without objective markers (Sherr & Shevell, 2012).

Challenging behavior is the most frequent reason for psychiatric referral and residential placement of individuals with developmental delay (Day, 1984). Challenging behaviors may include self-injurious, aggressive, destructive, impulsive, tantrum-like, being verbally or physically abusive, and sexually explicit or inappropriate behaviors. These behaviors may also be seen in conduct disorder and antisocial personality disorder (Marco, 2009a). The prevalence of challenging behaviors varies significantly depending on the definition of these behaviors and the ascertainment methodology used, with rates varying from 7 to 67% (Borthwick-Duffy, 1994; Campbell & Malone, 1991). Furthermore, the severity of the challenging behaviors appears to be inversely related to IQ (Borthwick-Duffy, 1994). Stressful life events (e.g., moving to a new home) can be associated with significant deterioration in behavior. Self-injurious behaviors have been found to be inversely associated with communication functions (Hubert-Williams & Hastings, 2008).

Both FXS and FAS tend to have disruptive behaviors related to poor self-regulation and impulse control, leading to decreased attention, hyperactivity, stereotypies, and disruptive behaviors both at school and at home (Mattson & Riley, 2000; Roebuck & Mattson, 1999). Self-injurious behaviors are characteristic of some rare genetic and metabolic disorder such as Lesch-Nyhan, Rett, and Cornelia de Lange but can also be seen in more common etiologies such as FXS, traumatic brain injury, and severe hypoxic injury (Marco, 2009a).

A multidisciplinary team should be involved, and a medical evaluation should focus on the identification of possible reversible illnesses that may cause or exacerbate challenging behaviors in a child with a lack of communication skills (e.g., intercurrent or chronic infections, allergies, medication side effects, dental caries, headache, gastroesophageal reflux, metabolic derangements, or dermatologic conditions) (Marco, 2009a).

Psychiatric disorders occur in 35–49% of children with ID (Dekker & Koot, 2003; Oeseburg, Jansen, Groothoff, Dijkstra, & Reijneveld, 2010b; Wallander et al., 2006). Multiple screening tools and checklists have been developed to facilitate recognition of specific psychiatric symptoms and include the Developmental Behaviour Checklist for Pediatrics (DBC-P) (Clarke, Tonge, & Einfeld, 2003) and the Children's Depression Inventory (Meins, 1993). Common examples of instruments used in adults include the Psychiatric Assessment Schedule for Adult with Developmental Disability checklist (PAS-ADD checklist) and the Developmental Behaviour checklist for adults (DBC-A) (Mohr, Tonge, & Einfeld, 2005). In more severe neurodevelopmental disorders, the DSM-5 criteria may be less applicable, and reasonable clinical inference may be required for diagnosis.

In a population-based study of British children and adolescents, 36% of children with ID had a psychiatric diagnosis compared to 8% among children without ID. Of these children with ID, 11.5% had anxiety disorders, 12% had emotional disorders, 8% had ADHD, and 20.5% had conduct disorders [most commonly oppositional defiant disorder (11%)]. Exposure to social disadvantage was also shown to be associated with an increased prevalence of psychiatric disorders (Emerson & Hatton, 2007). Another study found that anxiety was seen in up to 23% of children with developmental disability, which is significantly higher than the prevalence of 2–5% in typically developing children (Tonge et al., 1996). ADHD has a similar presentation among adolescents with and without ID except children

with ADHD and mild ID appear to have more conduct problems (Ahuja, Martin, Langley, & Thapar, 2013; Neece, Baker, Crnic, & Blacher, 2013). The majority of these individuals have more than one psychiatric disorder (Bakken et al., 2010). Another population-based study showed that 40% of children with ID had a psychiatric diagnosis and less than one third of the children with a psychiatric disorder received mental health-care services (Dekker & Koot, 2003). Individuals with intellectual disability are also at risk for developing suicidal thoughts and behaviors that may lead to suicide (Ludi et al., 2012).

Neurological Comorbidities

Neurological comorbidities are frequent in children with ID. A study in Sweden found that more than half of children with severe ID had one or more associated neurological conditions including: epilepsy (15–49%), cerebral palsy (18%), hearing impairment (7.5%), or vision impairment (12%) (Lundvall, Rajaei, Erlandson, & Kyllerman, 2012; Besag, 2002).

Children with developmental delays are at a higher risk to have vision and/or hearing impairments. Estimates of vision impairment or other visual disorders range from 13% up to 50%, and one series of patients found significant hearing impairments in about 18% of children (Shevell et al., 2003). These impairments may be congenital or acquired in origin. Vigilant and early hearing and vision screening in this population should be a standard of clinical practice, as early identification and treatment of sensory impairments can improve overall outcome for these individuals (Yoshinaga-Itano, 2003).

Several genetic disorders also place individuals at higher risk for epilepsy, as do acquired etiologies such as hypoxic ischemic encephalopathy or neonatal meningitis. It has been estimated that 20–30% of the patients with childhood epilepsy have ID (Besag, 2002; Camfield & Camfield, 2007; Wakamoto, Nagao, Hayashi, & Morimoto, 2000), approximately 50% of which were severe/profound (Camfield & Camfield, 2007). The long-term effects on cognition from seizures and the modulation of this by seizure control remains an area of interest in research.

Other Medical and Physical Comorbidities

Disordered sleep (especially insufficient sleep) is a very common problem in individuals with developmental disabilities and is typically complex and difficult to adequately control. Disordered sleep is so prevalent that it is included in the diagnostic criteria in several genetic syndromes associated with ID (e.g., Rett syndrome, Angelman syndrome) (Didden & Sigafoos, 2001; Grigg-Damberger & Ralls, 2013). Affected individuals have increased aggression, irritability, hyperactivity and other behavior problems. Insomnia in neurodevelopmental disorders is multifactorial and may be due to poor sleep hygiene, negative associations, and a lack of limit setting. Furthermore, abnormalities in genetic and/or epigenetic regulation of sleep and wakefulness may predispose to insomnia. Many children with ID/GDD also have hypotonia or extensive central nervous system abnormalities, and consequently central and obstructive sleep apnea is a common complication and can be diagnosed with polysomnography. A sleep history can be informative and should include sleep hygiene, nighttime awakenings, caffeine intake, and daytime behavior and can help guide behavioral treatment strategies (Didden & Sigafoos, 2001; Grigg-Damberger & Ralls, 2013; Harvey & Kennedy, 2002). Melatonin has been shown to be effective for individuals with ID/GDD and sleep disturbance and is well tolerated (Coppola et al., 2004; Gringras et al., 2012).

Difficulties with nutritional intake, undernutrition, gut motility, growth failure, and osteopenia are common in individuals with ID/GDD. Abnormal esophageal sphincter tone and oral motor incoordination are major factors leading to undernutrition, gastroesophageal reflux, and frequent aspirations. Moreover, gastroesophageal reflux may cause pain and irritability that can cause additional management issues. The placement of a percutaneous endoscopic gastrostomy may be necessary if oral intake is insuffi-

cient for growth and adequate hydration, the amount of time to feed the child is excessive due to swallowing dysfunction, or if there is an aspiration risk. Enteric feeding may improve quality of life for the individual, improve the ease of feeding the child and the familial stress involved with meals and feeding (Marchand & Motil, 2006; Marco, 2009b).

Course and Developmental Challenges

Intellectual disability has onset in the developmental period. All criteria for ID (including criterion C) must be fulfilled by history of current presentation. Several genetic syndromes are associated with variable degrees of ID and may have characteristic dysmorphic features which lead to a suspicion of ID during infancy before developmental delays become apparent (e.g., Down syndrome) (Walker & Johnson, 2006). Genetic syndromes can also have characteristic behavioral phenotypes. Some examples include Williams syndrome personality traits which have been described as unusually friendly and sociable, and they may also have an exceptional musicality (Meyer-Lindenberg, Mervis, & Faith Berman, 2006). Lesch-Nyhan syndrome presents with hypotonia and delayed motor skills usually evident by age 3–6 months, and almost all affected individuals eventually develop persistent self-injurious behavior (biting the fingers, hands, lips, and cheeks; banging the head or limbs) which is a hallmark of the disease (Nyhan, O'Neill, Jinnah, & Harris, 2000 Sep 25 [Updated 2014 May 15]). Untreated phenylketonuria presents with progressive intellectual disability, secondary microcephaly, and a "musty" body odor. Even treated individuals who strictly adhere to the recommended low-phenylalanine diet may develop cognitive impairments (Anderson et al., 2007; Moyle, Fox, Arthur, Bynevelt, & Burnett, 2007).

Some children under 5 years of age present with deficits that meet criteria for global developmental delay, with evidence of significant delays in two or more developmental domains, and they will eventually meet criteria for intellectual disability. However, having a diagnosis of GDD is not necessarily associated with cognitive impairment or a diagnosis of ID. A cohort of 93 children with GDD showed average intelligence in 20% of the cohort (Riou, Ghosh, Francoeur, & Shevell, 2009).

Etiology identified may also predict the course of ID. ID is generally lifelong and non-progressive, but there may also be a deteriorating course, or a gradual resolving course. In certain genetic disorders (e.g., Rett syndrome), there are periods of worsening, followed by stabilization (Shahbazian & Zoghbi, 2001), whereas in others there is a progressive worsening (e.g., Sanfilippo syndrome due to progressive accumulation of mucopolysaccharides in the brain and other organs) (Valstar, Ruijter, van Diggelen, Poorthuis, & Wijburg, 2008). Medical conditions or co-occurring sensory impairments may also affect the course and severity level. Measures of cognitive function during infancy are known to be poor predictors of later IQ, although there is stability and continuity over time in specific subdomains of function on a neurodevelopmental screening test. There is evidence for brain plasticity, and the diagnosis may change over time due to early and ongoing interventions that may improve adaptive functioning, and the diagnosis of ID may indeed occasionally no longer be applicable. Consequently, it is advised that for infants and young children, the definitive diagnosis of ID should be delayed until after an appropriate course of intervention is provided (Aylward, 2004; Hack et al., 2005; Illingworth & Birch, 1959; Johnston, 2009; Yang, Yong, Hsu, & Lung, 2011). Interventions may help acquire important activities of daily living, and adaptive life and vocational skills. Diagnostic assessments can be used to aid in the determination of whether the improved adaptive skills during or following an intervention are due to a stable, generalized new skill acquisition (in which case the diagnosis of intellectual disability may no longer be appropriate) or whether the improvement is dependent on the presence of additional supports and ongoing interventions (in which case the diagnosis of intellectual disability may still be appropriate) (APA, 2013a).

Evaluation

Neurodevelopmental, Intellectual and Behavioral Assessment

A comprehensive evaluation includes an assessment of intellectual capacity and adaptive functioning (APA, 2013a). Frequently the evaluation takes more than one encounter and often requires the combination of observations and data from a multidisciplinary team. Several tools have been developed to measure development, intelligence/cognition, and neuropsychological development, as well as indirect functional ratings that can be reported by the parent or caregiver.

Developmental delay is reserved for children less than 5 years of age and can be diagnosed with the Bayley Scales of Infant Development (Bayley, 2006). Other broad developmental standardized tests include the Battelle Developmental Inventory (Newborg, Stock, Wnek, Guidubaldi, & Svinicki, 1984) and the Denver Developmental Screening Test (second edition) (Frankenburg, Dodds, Archer, Shapiro, & Bresnick, 1992). Often, domain-specific measures are individually applied and used to form an overall assessment. Some examples of domain-specific developmental measures include the following groups:

1. *Motor profile*: Alberta Infant Motor Scale (AIMS) (Piper & Darrah, 1994) or the Peabody Developmental Motor Scales (PDMS) (Folio & Dubose, 1974)
2. *Language skills*: Peabody Picture Vocabulary Test – Revised (PPVT-R) (Dunn & Dunn, 1997) or the Expressive One Word Picture Vocabulary Test – Revised (EOWPVT-R) (Gardner, 1990)
3. *Behavior and activities of daily living*: Vineland Adaptive Behavior Scale (VABS) (Sparrow, Balla, & Cicchette, 1984) or Pediatric Evaluation of Disability Inventory (PEDI) (Haley, 1992)
4. *Participation and quality of life*: Children's Assessment of Participation and Enjoyment (King et al., 2004), Assessment of Life Habits (Fougeyrollas & Noreau, 2002), the Peds QL (Varni, Seid, & Kurtin, 2001), and the KIDSCREEN (Erhart, Ravens-Sieberer, Dickinson, Colver, & the European SPARCLE and KIDSCREEN Groups, 2009)

ID is diagnosed with the Wechsler preschool and primary scale of intelligence (WPPSI-IV) and Wechsler Intelligence Scale for children (WISC-IV) (Wechsler, 1967, 1991, 2003b). WPPSI-IV has been standardized to age 2 years 6 months to 7 years 7 months, but has recognized limitations in interpretability (Sattler, 1982). WISC-IV assesses cognitive abilities in children age 6 to 16 years 11 months. Other measures include the Stanford-Binet IV which is used for children and adults (Thorndike, Hagen, & Sattler, 1986), among others.

Adaptive function is assessed with the Vineland Adaptive Behavior Scale (VABS) (Sparrow et al., 1984). There are acceptable alternatives which include the AAMR Adaptive Behavior Scale (ABS) (Lembert, Nihira, & Leland, 1993), the Scales of Independent Behavior – Revised (SIB-R) (Woodcock, Weatherman, & Hill, 1996), the Comprehensive Test of Adaptive Behavior – Revised (CTAB-R) (Adams, 1999), and the Adaptive Behavior Assessment System (ABAS) (Harrison & Oakland, 2000).

Often the diagnosis of GDD and ID may be initially formulated or, less frequently, entirely based on clinical judgment, rather than on formal standardized tests. Such clinical judgment must be based on extensive direct experience with individuals with GDD or ID and should be validated by (1) direct observation, (2) information from reliable third-party informants, (3) input from an interdisciplinary team skilled in multidimensional standardized assessments, and (4) repeated observations of an individual over time. Clinical judgment may be necessary because of various social, cultural, and linguistic contexts or because of unavailability, inappropriateness, or delay in the administration of standardized assessment procedures (Sherr & Shevell, 2012).

History and Physical Examination

There are multiple objectives in the medical evaluation, and these include the following elements:

1. Confirming and classifying the precise neurodevelopmental disability
2. Through history, physical examination, and selective laboratory testing, searching for a possible underlying etiology
3. Arranging for appropriate needed supports and rehabilitation service interventions
4. Counseling the family regarding the implications of the diagnosis from individual and familial perspectives, including a discussion of possible recurrence risks and possible outcomes
5. Identifying and managing associated medical or behavioral conditions (e.g., seizure disorders, attention difficulties, sleep disturbances, spasticity, behavioral disorders) (Shevell, 2006, 2009)

Careful clinical and family histories with detailed physical and neurological examination are central to the initial evaluation. During the evaluation, the main etiologic categories to keep in mind are cerebral dysgenesis, genetic syndrome/chromosomal abnormalities, intrapartum asphyxia, antenatal toxin exposure, or psychosocial deprivation (Shevell, 2009; Srour et al., 2006). A comprehensive three-generational family history should include the possibility of parental consanguinity as well as any familial neonatal or infantile deaths or recurrent maternal pregnancy losses. The details regarding the prenatal, postnatal, and neonatal period should be established including any suggestions of difficulties during the delivery process and should include birth weight, Apgar scores, head circumference, duration of postnatal hospital stay, and if any difficulties in the newborn nursery (e.g., feeding difficulties, seizures). Developmental progress in each of the domains (i.e., motor, language, cognitive skills, activities of daily living) should be established including the possible loss of skills or developmental regression as this would require a different and more urgent approach to the evaluation and follow-up (Sherr & Shevell, 2012; Shevell, 2009). Clinical features that can help predict a successful etiologic diagnosis include: female gender, abnormal prenatal/perinatal history, microcephaly, abnormal neurological examination, dysmorphic features and an absence of autistic features (Srour et al., 2006).

The physician should inquire about the current social situation of the child and family including items such as socioeconomic status, parental educational attainment, marital status and home living arrangements, child custody and existing support services. It should also be determined whether the patient has access to appropriate rehabilitation services (e.g., occupational therapy, physiotherapy, speech therapy, special education, psychology) and whether relevant evaluations and laboratory investigations have been previously conducted (Sherr & Shevell, 2012).

The physical examination is an integral part of the diagnostic evaluation of the delayed child as it may suggest a specific syndrome. Ideally, the examination begins with careful observation of the child as the history is being taken. The formal examination may be a challenge because of the inherent features of delayed children, such as short attention span and limitations in understanding and cooperation. It is useful to have a play area available (with appropriate toys, including paper, crayons, dolls, and representational toys), so the examiner may observe developmental skills, dexterity, inquisitiveness, behavior, and interaction with the surroundings and others in a nonintrusive fashion. Affective, temperamental, and regulatory patterns and any unusual behaviors should be documented (e.g., child's adaptation to new environments and people, state transitions such as sleep/wake and eating behaviors). The child-parent interaction, as well as the child's solitary play and social interaction with others, should be observed. During the physical examination, particular attention should be paid to possible dysmorphic features, stigmata of neurocutaneous disorders, palpable hepatosplenomegaly or coarsening of facies (suggestive of storage disorders), head circumference (microce-

phalic or macrocephalic measurements and if there is a change in percentile values over time), lateralizing features, and weakness or tone changes (Sherr & Shevell, 2012; Shevell, 2009; Tirosh & Jaffe, 2011).

Diagnostic Investigations and Laboratory Testing

The AAN practice parameter and evidence report for global developmental delay (Michelson et al., 2011; Shevell et al., 2003) provide a framework for an evaluation. Due to genetic advances, the evidence report focused solely on the diagnostic testing of genetic and metabolic disorders. The practice parameter and evidence report incorporate a combination of broad screening tools and disease-specific testing based on a heightened pretest probability, given identifying clinical features (physical, neurologic, ophthalmologic examination, and basic laboratory screening tests). Correctly applied, each has a reasonable pretest probability (>1%) of diagnosis. The algorithm begins with a complete clinical assessment. For those patients in whom a specific diagnosis is considered, targeted testing is recommended early in the evaluation (e.g., an MRI for an asymmetric physical examination, methylation testing for Angelman syndrome, or testing for Prader-Willi syndrome and bone age determination for Sotos syndrome in the setting of large somatic features). For the remaining patients, a stepwise approach was recommended.

Microarray is the current genetic test with the highest diagnostic yield in children with unexplained GDD/ID, and there is consensus that aCGH should be considered the first-line cytogenetic test, preferred over StFISH testing and karyotyping (Michelson et al., 2011). This recommendation is based on many recent studies showing the enhanced utility of aCGH to detect clinically relevant chromosomal changes, and it is also recommended by the 2010 American College of Medical Genetics practice guidelines for children with multiple anomalies or for cases of nonsyndromic GDD/ID or ASD. Karyotyping should be reserved for patients that have signs of a specific chromosomal syndrome (e.g., Down syndrome), a family history of a chromosomal rearrangement, or a parent with a history of multiple miscarriages (Manning & Hudgins, 2010; Michelson et al., 2011; Miller et al., 2010; Sherr et al., 2013).

There are some notable limitations of aCGH, including that only unbalanced copy number changes are detected, and consequently arrays cannot identify balanced inversions/insertions or reciprocal translocations. Also, because of the overall resolution, aCGH will miss low-level mosaicism (typically <20%), polyploidy, rearrangements in repeat sequences, and point mutations or small insertions or deletions in single genes (Michelson et al., 2011; Sherr et al., 2013). Additionally, "variants of unknown significance" require thoughtful interpretation. These are variants that are rarely seen in the general population or are unique to a family (frequency < 0.001) and are not a priori known to be associated with disease. The first step is to establish whether a documented copy number change is de novo or familial, although it can still be difficult to try to determine whether rare CNVs are pathogenic or benign. As additional data is collected from large populations, the establishment of comprehensive, computerized, publicly available genotype-phenotype databases aid in the rapid interpretation of the pathogenicity of these less common copy number changes. Two public databases include the database of genomic structural variation (dbVar) and ClinVar, which are maintained by the National Center for Biotechnology Information (Sherr et al., 2013).

FMR1 testing shows full expansion in at least 2% of patients with mild-to-moderate GDD/ID and is recommended to evaluate children with mild ID of both genders. Males with a history strongly suggestive of X-linked inheritance may be considered for testing of one or more individual XLID genes or for screening of the entire X-chromosome (Michelson et al., 2011). Next-generation sequencing panels are available for XLID and can screen for a large number of genes (Sherr et al., 2013). MeCP2 testing (for Rett syndrome) is recommended for moderate to severely affected females, regardless of whether

the specific clinical features of Rett syndrome are present (diagnostic yield of 1.5%) (Michelson et al., 2011).

However, there are rapid advances in the field, and newly evolving diagnostic tools are appearing in genetic testing, including WES and whole genome sequencing (WGS) (Sherr et al., 2013). As the cost of WES has dropped substantially in the past few years, and the ability to analyze the data has improved, WES may soon become standard testing similar to microarray currently in children with ID/GDD (Challis et al., 2012; Sherr et al., 2013). However, there are many ethical challenges as WES may yield a surplus of results that are unrelated to the child's developmental disability but may be of importance to the patient in other ways (Tucker, Marra, & Friedman, 2009).

If the first-line tests are unrevealing, high-resolution MRI is then recommended (with concurrent single proton spectroscopy, where that is available) and is preferable to computed tomography, particularly if there are findings on examination. Indications for MRI include macrocephaly or microcephaly, asymmetric neurologic findings, intractable epilepsy or focal seizures, abnormal movements (e.g., dystonia, chorea, or other extrapyramidal findings), hypotonia or long tract signs, facial stigmata associated with developmental brain abnormalities, and a history of a progressive neurologic disorder (Mefford et al., 2012). Proton MR spectroscopy assesses brain chemistry by measuring the unique resonance frequencies of several molecules in the brain. This approach has been useful for detecting changes in cerebral lactate in mitochondrial disorders and for observing the absence of creatine in disorders of creatine deficiency (Stromberger, Bodamer, & Stockler-Ipsiroglu, 2003).

Studies suggest that MRI is useful for detecting abnormalities in 48.6–65.5% of children with GDD, with the chance of detecting an abnormality increasing if physical abnormalities, particularly cerebral palsy, are present (Shevell et al., 2003). The yield is higher if neuroimaging studies are performed on an indicated basis rather than a screening basis (Shevell et al., 2003; van Karnebeek et al., 2005). Hart et al. found that 44% of patients with isolated GDD had an abnormal MRI, whereas 55.3% of children with GDD and additional symptoms had abnormal MRI. Half of each group had specific abnormalities (e.g., brain malformations, hypoxic ischemic encephalopathy), whereas nonspecific abnormalities (e.g., reduced volume of white matter, delayed myelination) were noted in the other half (Hart et al., 2011). More abnormalities are found on MRI in children with a moderate to profound intellectual disability than those with a borderline to mild disability (van Karnebeek et al., 2005).

If this approach is not diagnostic, comprehensive metabolic testing is then recommended. Metabolic testing should be performed if there are symptoms or historical features suggestive of inborn errors of metabolism such as parental consanguinity, prior affected family members, unexplained death or fetal demise, failure to thrive, multiple organ system dysfunction, dietary selectivity, unusual odors, hearing loss, developmental regression, episodic decompensation (including seizures or encephalopathy), as well as any suggestive physical findings (e.g., hepatosplenomegaly, coarse facial features) or neuroimaging findings (abnormal myelination, striatal necrosis) (Michelson et al., 2011; Shevell et al., 2003). Routine screening for inborn errors of metabolism in GDD/ID has a yield between 0.2% and 4.6%, depending on the presence of clinical indicators and the range of testing performed. Testing for congenital disorders of glycosylation (CDGs) has a yield of up to 1.4%, and testing for creatine synthesis and transport disorders has a yield of up to 2.8% (Michelson et al., 2011). The yield may increase to about 14% when a stepwise approach is used (based on physical, neurologic and ophthalmologic examination and basic laboratory screening tests) (Shevell et al., 2003).

Electroencephalogram (EEG) is not recommended as part of the initial evaluation unless there is suggestion on history of seizures/paroxysmal behaviors, of a specific epilepsy syndrome, or clinical features that suggest a possible acquired epileptic aphasia (e.g., Landau-Kleffner syndrome or electrical status epilepticus during slow-wave sleep), such as objective evidence of language regression or behavioral disturbances

(Sherr & Shevell, 2012; Shevell, 2009; Shevell et al., 2003).

Lead testing should only be done in those children with identifiable risk factors for excessive lead exposure. Similarly, testing for thyroid hormone status should target those in whom newborn screening was not undertaken or in those with specific systemic features of hypothyroidism (Shevell, 2009).

There is a high frequency of primary sensory impairments in this population; thus, all children with GDD should undergo standardized hearing and vision assessment (Shevell, 2009; Shevell et al., 2003). The patient should also be screened for co-occurring mental, emotional, and behavior disorders (APA, 2013a).

Treatment

A multidisciplinary team is usually required in the management of a child with GDD/ID and may include as appropriate a social worker, occupational therapist, physiotherapist, speech language pathologist, psychologist, psychiatrist, pediatric neurologist, developmental pediatrician and medical geneticist. Medical consultation for the child's general health needs to be obtained as well (Shevell, 2009; van Karnebeek et al., 2014). Treatment focuses on actualized individualized rehabilitation and education plans, targeting specific interventions for treatable disorders and treating comorbidities (e.g., epilepsy, sleep).

Early intervention has proven to be beneficial in randomized clinical trials in children with GDD/ID, including children with genetic causes of disability, suggesting that the early diagnosis of a child with global delay may improve outcome (Einfeld, Tonge, & Clarke, 2013; Guralnick, 1998, 2005; Landry, Smith, Swank, & Guttentag, 2008; Ludwig & Phillips, 2008; Thomaidis, Kaderoglou, Stefou, Damianou, & Bakoula, 2000). Head Start is the US government's major early childhood education program and has shown to produce short-term effects and possibly long-term benefits as well (Ludwig & Phillips, 2008). The rationale for early intervention in children with developmental disabilities is associated with the concept of brain plasticity. Beyond genetic programing, there are experience-dependent changes that individualize and shape development. Repetitive stimulation of synapses can cause long-term potentiation or long-term depression of neurotransmission. These changes are associated with physical changes in dendritic spines and neuronal circuits. Mechanisms for neuronal plasticity contribute to the developing brain's ability to acquire new information, change in response to environmental stimulation, and recovery from injury (Johnston, 2009).

Therapeutic modalities exist for certain metabolic disorders which can improve clinical outcomes and include supplements, specific dietary treatments, substrate inhibition, enzyme replacement therapies or even stem cell transplantation in rare instances (Sherr et al., 2013; van Karnebeek et al., 2012).

There is also relatively high use of complementary and alternative medicine in children with neurodevelopmental disorders, and these should be discussed with families including potential side effects or interactions and our current knowledge of safety and efficacy of these therapies (Akins, Krakowiak, Angkustsiri, Hertz-Picciotto, & Hansen, 2014; Valicenti-McDermott et al., 2014).

Treatment of Behavioral and Psychiatric Disorders

First-line interventions are behavioral, and medical therapies should be used along with non-medication-based management plans. Individuals with ID or neurological impairments are more sensitive to medication side effects, and thus they should be used with caution including lower dosage and slower dosage increases than in the general population. Several studies support medical therapies for the treatment of psychiatric conditions in the intellectually disabled population. The major categories of medications used in challenging behaviors modulate dopamine, serotonin, and opiate availability (Handen & Gilchrist, 2006; Marco, 2009a). The newer second-generation neuroleptics (especially risperidone)

have been effective for behavioral disorders, particularly aggression and impulsivity (Croonenberghs, Fegert, Findling, de Smedt, & Van Dongen, 2005; Snyder et al., 2002). Only risperidone has shown significant efficacy in randomized placebo-control trials in children with ID and borderline intelligence (Unwin & Shoumitro, 2011). Others such as clozapine, quetiapine, ziprasidone and aripiprazole have also been used to treat aggression, self-mutilation, and psychiatric syndromes. Clozapine's use is limited due to severe side effects such as agranulocytosis (Handen & Gilchrist, 2006). Side effects of the narcoleptic medications include mostly weight gain and somnolence, but extrapyramidal symptoms, headache, and seizure exacerbation are also seen. Moreover, laboratory screening is required for many (Handen & Gilchrist, 2006; Unwin & Shoumitro, 2011). There is some evidence that naltrexone may be beneficial for self-injurious behaviors (Symons, Thompson, & Rodriguez, 2004).

ADHD is a common comorbidity in children and adolescents with ID. Methylphenidate is available in short- and long-acting forms and is effective for patients with ID and ADHD, although the response rates are lower (45–66%) than in typically developing children (77% response rates) (Aman, Buican, & Arnold, 2003; Handen & Gilchrist, 2006; Lipkin, 2013; Pearson et al., 2004). The prevalence of ADHD symptoms in children with FXS is higher than other genetic conditions or individuals with nonspecific intellectual disability. Methylphenidate at usual doses showed a positive response in 70% of FXS patients in controlled trials (Hagerman et al., 2009; Hagerman, Murphy, & Wittenberger, 1988). Children and adolescents with ID may be at greater risk of experiencing side effects with methylphenidate (Handen & Gilchrist, 2006). Alpha agonists such as clonidine and guanfacine are also used for impulsivity, hyperactivity, and inattention. Since stimulants may induce irritability and other behavioral problems in children less than 5 years of age, alpha agonists seem to be a good option for younger children or neurologically more affected children who have adverse effects with stimulants (Agarwal, Sitholey, Kumar, & Prasad, 2001; Hagerman et al., 2009). Clonidine can also be helpful for children who have sleep disturbances (Ingrassia & Turk, 2005). Selective serotonin reuptake inhibitors (SSRIs) have been shown to be helpful for anxiety, social anxiety disorder, depression, obsessive-compulsive disorder and stereotypic or self-injurious behaviors (Handen & Gilchrist, 2006).

Pharmacological Treatment for Cognitive Impairment

Advances in genetic techniques are leading to the identification of many more genes that either act directly or are part of common pathways in emerging networks. Although there is significant genetic heterogeneity, it seems there are common pathological patterns in ID, and the failure of appropriate signaling between neurons across the synaptic junctions of dendritic spines is the central deficit in many cases of ID. A common molecular etiology could be targeted even in a group of genetically heterogeneous individuals with ID. The goal in the understanding of the molecular pathogenesis of ID is the development of specific mechanism-based therapy (Ropers, 2010; van Bokhoven, 2011). Consequently, obtaining a proper genetic diagnosis has the possibility of directing patients toward targeted treatments in the future. Finding a genetic diagnosis can also positively influence current pharmacological options or disease surveillance. For example, if an SCN1A mutation or a GLUT1 defect was found, this would lead to specific changes in interventions that would improve seizure management and developmental outcomes (Sherr et al., 2013).

The best example of this is FXS. The fragile X mental retardation protein (FMRP) is the product of the fragile X gene (FMR1). FMRP regulates dendritic growth by regulating local protein translation through inhibition of metabotropic glutamate receptor 5 (mGLUR5 receptors). FMRP is thus critical for synaptic maintenance and plasticity. Mutation of FMRP results in unimpeded mGluR5 activity, which causes aberrant dendritic development and mis-signaling,

and leads clinically to ID, autism, and psychopathology. This pathway contains many potential therapeutic targets (Dictenberg, Swanger, Antar, Singer, & Bassell, 2008; Muddashetty, Kelic, Gross, Xu, & Bassell, 2007; Picker & Walsh, 2013; Zalfa et al., 2003). For instance, GABAergic activity can be increased. Clinical trials showed that Arbaclofen, a (GABA(B)) agonist, may have the potential to improve social function and behavior in fragile X (Berry-Kravis et al., 2012). Additionally, studies showed that the memory defects and social interaction defects observed in fragile X flies and mouse models could be reversed using mGluR5 antagonists (van Bokhoven, 2011). There are several mGluR5-specific antagonist trials (AFQ056, RO4917523, and STX107) underway, with the goal of replacing the inhibitory effect of the missing FMRP activity (Jacquemont et al., 2011; Picker & Walsh, 2013). A phase I open label trial of the mGluR5 inhibitor fenobam suggested promising efficacy based on a single dose (Berry-Kravis et al., 2009).

Interestingly, many of the pathways found to be involved in ID have also been studied for their role in tumors and other disorders, and a wide variety of medications have been identified that can correct the effects of disrupted agents within these pathways. Consequently, possible uses for already-approved medications are increasingly being tested for their ability to rescue phenotypic features in animal models and in ID patients (van Bokhoven, 2011). A modulator of protein synthesis related to the ERK pathway, lovastatin (widely prescribed for the treatment of high cholesterol) was shown to reverse the exaggerated long-term depression (LTD) and epilepsy in fragile X mice (Osterweil et al., 2013). Additionally, the antibiotic minocycline, a metalloproteinase inhibitor, appeared in a double-blind study to have some efficacy (Leigh et al., 2013). Valproic acid, a common antiepileptic medication with histone deacetylation (HDAC) inhibitory activity, has been tested for its potential to reactivate *FMR1* gene expression, and preliminary results suggested a beneficial effect on the symptoms of ADHD in young patients with FXS (Torrioli et al., 2010). Likewise, lithium and baclofen use has shown some improved cognitive performance in a Down syndrome mouse model (Contestabile et al., 2013; Kleschevnikov et al., 2012).

Trisomy 21/Down syndrome is the most common genetic cause of ID. Although there is duplication of an entire chromosome, it is believed that only a small number of genes and other genetic elements are involved in the phenotype of Down syndrome. There are several new promising studies on pharmacological agents capable of rescuing learning and memory deficits seen in mouse models of Down syndrome. Some examples include epigallocatechin gallate (EGCG) which is a polyphenol that is a fairly specific DYRK1A inhibitor. DYRK1A is located on chromosome 21 and is overexpressed in Down syndrome and has been shown to be associated with neurofibrillary tangles and splicing regulation (Costa & Scott-McKean, 2013). Vitamin E is currently in trials to see if it will slow the cognitive decline of older adults with Down syndrome who develop early-onset Alzheimer disease. Mice models with vitamin E supplementation have shown improvements in memory. Memantine (an NMDA receptor antagonist) is currently one of the most studied potential pharmacotherapies for Down syndrome in a preclinical setting. A study using memantine suggested limited cognitive improvement in verbal memory in adults with Down syndrome (Boada et al., 2012; Costa & Scott-McKean, 2013; Picker & Walsh, 2013).

Another common cause of ID/ASD is tuberous sclerosis (TS). TS is caused by mutations in either TSC1 or TSC2, which encode proteins that form a complex that inhibits activation of mammalian target of rapamycin (mTOR), which is a potential target for treatment. Everolimus, an inhibitor of mTOR, is currently in clinical trials to assess its role in improving the neurocognitive function of TS patients with ID and ASD. The mTOR pathway interweaves with the fragile X pathway, and it is possible that these treatments may help at least some other ID/autistic disorders in which the pathway appears to be indirectly disrupted (Ehninger et al., 2008; Picker & Walsh, 2013).

Prognosis and Outcome

The clinical impact of ID depends on many factors, including level of severity, access to resources and environmental conditions. The vast majority of children with intellectual disability or GDD remain at home with caregivers. Family support to ensure family health and well-being should be provided, especially in the context of severe delay or intellectual disability, such as short-term respite care. A social worker can also help to ensure that access to available governmental supplemental financial resources is facilitated. In the United States, federal law mandates the provision of early education and rehabilitation services for preschool children with a developmental disability and appropriate educational services for school-age children, regardless of the severity of the ID (Sherr & Shevell, 2012). Better developmental outcomes are found in children with more positive family support and provided home or foster care rather than institutional or residential care (Shevell, Majnemer, Platt, Webster, & Birnbaum, 2005a). Siblings of the delayed child more often feel lonely and are prone to more frequent peer problems and behavioral disturbances (Bagenholm & Gillberg, 1991).

Global Developmental Delay

Studies have suggested that early childhood developmental delays are correlated with later academic problems during the school years (Montgomery, 1988; Shapiro et al., 1990). Early motor and cognitive delays have been associated with significant reading difficulties at school age (Shapiro et al., 1990), and early perceptual motor difficulties precede poor performance on early mathematic concepts (Feder & Kerr, 1996).

Longitudinal prospective studies have also demonstrated persisting functional and developmental disabilities and suggest a prognostic validity to the diagnostic label of global developmental delay (Shevell et al., 2005a). The degree of initial delay predicted later functional disabilities, but not eventual developmental outcome. Higher functional communication and socialization skills were associated with maternal employment and paternal post-secondary education, which reflected a higher socioeconomic status, and suggested that these parents more effectively advocated for access to needed resources (Shevell et al., 2005a; Shevell, Majnemer, Platt, Webster, & Birnbaum, 2005b).

Mild to Moderate Intellectual Disability

Longitudinal studies suggest continued intellectual development in those with mild or moderate delay (Eyman & Widaman, 1987). Most problems begin at school age (especially with reading, writing and timed tests) and with interpersonal relationship skills with peers. With adequate training, most individuals with mild ID achieve full independence in most domains of function but still have problems with the adaptive integrative domain (Pratt & Greydanus, 2007). Practically, the focus of interventions should be on skills that assist in promoting functional capacity, especially with respect to obtaining independence in activities of daily living (i.e., autonomy). Life skills and employment training are important goals for those with mild or moderate intellectual disability and appear to be integral to achieving a sense of self-worth and self-esteem (Sherr & Shevell, 2012). Important factors in the successful transition to adult life include parental involvement in transition planning, an individual's educational achievements, and the ability to acquire basic living skills (Reiter & Palnizky, 1996).

Individuals with mild ID may develop major psychosocial problems in adolescence when abstract thinking, problem solving, and critical thinking, and developing the ability to engage in sustained employment and mutually healthy intimate relationships becomes the main objectives of development. Individuals with mild-to-moderate ID can successfully live independently as adults; however, the guidance, training, and support they receive as children and adults will determine the actual level of success an individual achieves (Pratt & Greydanus, 2007).

Individuals with moderate ID have more significant deficits in social communication and language expression and comprehension. They will need guidance and support throughout their lives. Semi-independent living conditions are usually required since regular reminders may be needed for individual care of personal needs (i.e., eating, hygiene). These individuals can develop simple friendships and a variety of recreational skills can be developed. They will need close supervision (supported or sheltered) in employment endeavors but can be very dependable if given tasks that require limited conceptual and communication skills, with appropriate training and support provided (APA, 2013a; Pratt & Greydanus, 2007). Individuals with jobs in the community have significantly higher levels of quality of life and self-esteem. Thus, although periodic and ongoing support is often needed for continued employment, it should be pursued (Kraemer, McIntyre, Blacher, & Taylor, 2003).

Severe to Profound Intellectual Disability

Longitudinal studies suggest an absence of continued improvement in intellectual development in those with severe or profound intellectual disability. The functional attainment by age 6 years in children with severe neurodevelopmental disability typically represents the functional attainment with respect to the ambulation, feeding, toileting, and self-hygiene they will achieve with substantially diminished chances for further functional improvement after that age (Strauss, Ashwal, Shavelle, & Eyman, 1997).

With a higher severity of ID, there are often marked motor impairments and associated deficits, and thus the degree of functioning decreases. These individuals will need care and supervision to perform all activities of daily living, and they will require supervision at all times. Ongoing support and teaching are required to participate in tasks at home and recreationally (APA, 2013a; Pratt & Greydanus, 2007).

Children with profound ID have severely limited understanding of language comprehension and expression and a resulting inability to understand instructions. They are often severely restricted in mobility, sensory impairments, and continence and require constant help and supervision in all aspects of daily physical care, health and safety. Those without severe physical impairments may be able to participate in simple daily tasks (e.g., carrying dishes to the table) (APA, 2013a; Pratt & Greydanus, 2007).

Life Expectancy

The life expectancy of a child with mild-to-moderate ID without severe and multiple disabilities or Down syndrome approximates the general population. Advances in medicine, health care, and nutrition, together with unimpeded access to care, have resulted in adults with ID living longer (Coppus, 2013). However, life expectancy is still negatively associated with the severity of ID. A recent population-based study in England found the median age at death for mild ID was 68 years, moderate ID 64 years, severe ID 59 years, and profound ID 46 years. The median age at death of those with mild intellectual disabilities was still substantially younger than the general population (Heslop, Fleming, Highton, Marriott, & Russ, 2014). There is also some evidence of a racial disparity in survival with the median age of death significantly lower in non-Caucasians with Down syndrome (Yang, Rasmussen, & Friedman, 2002).

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Language Disorder in Children

3

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Overview of Language Disorder and History in DSMs

Children with a language disorder have unusual difficulties understanding and/or using vocabulary or grammar in age-appropriate ways. Their language impairments cannot be attributed to underlying conditions such as intellectual disability, sensory disorders (hearing or vision), medical conditions, neurological disorders, or motor dysfunctions (American Psychiatric Association [APA], 2013). For example, children who are born with Down syndrome, severe hearing impairment, or fetal alcohol syndrome are likely to have problems learning and using language, but these children should receive diagnoses that are based on their primary disorder. Thus, the diagnosis of language disorder should be reserved for children whose receptive and/or expressive language skills are significantly below that which would be expected for their age in the absence of any other developmental disability that could cause the language difficulties.

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Language disorder occurs fairly commonly, affecting approximately 7% of preschool and school-age children (Tomblin et al., 1997). Children whose language-learning difficulties do not resolve by age 5 frequently have difficulties with social and academic language during the elementary school years (Tomblin, Zhang, Buckwalter, & O'Brien, 2003), and they are likely to continue to exhibit social, academic, and vocational difficulties well into the high school years (Johnson et al., 1999; Stothard, Snowling, Bishop, Chipchase, & Kaplan, 1998). Because reading and writing are language tasks (Kamhi & Catts, 1999), many school-age children with language disorders present literacy learning problems (Catts, Fey, Weismer, & Bridges, 2014). Terms such as developmental language disorders, specific language impairment, and primary language impairment are also used as diagnostic labels for unexplained developmental language problems (Bishop, 2014).

History of Language Disorder

Until the late 1950s, language disorders and speech disorders were collapsed into a single category of "speech disturbances" (DSM-I, 1952) or "other speech impediments" (ICD-7, 1957). The modern era of clinical research that focused on children who demonstrated significant language

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delays without co-occurring mental retardation, hearing impairment, or autism began in the 1960s, when researchers like Jon Eisenson at Stanford studied children he referred to as having developmental aphasia (Eisenson, 1966, 1968). About that same time, Mildred Berry and her colleagues at Texas Woman's University and the University of Wisconsin at Madison were studying children that they referred to as having idiopathic language retardation or neurogenic learning disabilities (Berry, 1969). Most researchers at the time assumed that such children had atypical neural organization, often referred to as "minimal brain dysfunctions" (Clements, 1966) that interfered with language learning.

The ICD-9 (1978) differentiated speech and language disorders, but both types of disorders were part of a single diagnostic category called "developmental speech/language disorders." It was not until the DSM-III in 1980 that children with language disorders had their own diagnostic category (developmental language disorder – expressive and receptive type) that was separate from a "developmental articulation disorder." The designation was further divided into developmental expressive language disorder and developmental receptive language disorder in the DSM-III-R (1987).

In both the DSM-III and the DSM-III-R, developmental language disorders were part of the category of specific developmental disorders that included developmental reading disorder (dyslexia), developmental arithmetic disorder, and mixed specific developmental disorder. The key to the diagnosis was that the child had a particular skill that was impaired relative to age expectations, but overall development was within the normal range. Grouping developmental reading disorder and developmental language disorder in the same category acknowledged the close relationships between these conditions.

In the DSM-5, communication disorders (including language disorder) are categorized as neurodevelopmental disorders along with intellectual disability, autism spectrum disorder, attention-deficit/hyperactivity disorder, specific learning disorder, and motor disorders. The disorders in this grouping have similar genetic risk factors, shared neural substrates, and similar clinical features such as cognitive processing problems, higher rates of comorbidity with each other, and a continuous course of development into adulthood (Andrews, Pine, Hobbs, Anderson, & Sunderland, 2009). It is interesting that in some respects, the current characterization of language disorder as a neurologically based disorder is similar in nature to the neurologically oriented characterization that was prevalent in the 1960s, albeit with a much stronger scientific base.

Current Diagnostic Criteria for Language Disorder

Language disorder is characterized by impairments in vocabulary, sentence structure, and discourse level communication (APA, 2013; American Speech-Language Hearing Association [ASHA], 1993). These difficulties should be apparent across multiple modalities including speaking, writing, and/or sign language (APA, 2013; ASHA, 1993). Language disorder affects receptive language, expressive language, or both. Receptive language relates to an individual's ability to comprehend linguistic information, whereas expressive language relates to an individual's ability to formulate and produce linguistic information. To be diagnosed with a language disorder, the child's language performance must be shown to be significantly below age expectations to the extent that it would be likely to interfere with communicative, social, academic, and/or vocational functioning. Finally, a language disorder must be present from early development and must not be attributable to intellectual impairment or global developmental delay. Language difficulties exist in the absence of or to a greater degree than could be explained by other sensory, motoric, medical, or neurological conditions that may be present (APA, 2013).

The DSM-5 does not list specific psychometric criteria for diagnosing language disorder. To date, the only epidemiologically derived diagnostic criterion comes from a series of longitudinal studies of kindergarten children with specific language impairment by Tomblin and his colleagues (Catts, Bridges, Little, & Tomblin, 2008; Fey, Catts, Proctor-Williams, Tomblin, & Zhang, 2004; Leonard et al., 2007; Tomblin et al., 2003). Tomblin, Records, and Zhang (1996) reported excellent sensitivity and specificity (values above .9) for a

standard score cutoff of 83 on a global language test that assessed receptive and expressive language skills in the areas of vocabulary, syntax, and discourse processes or for performance of −1.25 standard deviations or more below the mean on two or more composite scores representing language comprehension, language production, vocabulary skill, grammatical skill, and narrative skill. However, looking across a number of studies of children with language disorders, many investigators identify language disorder by performance that is −1 or more standard deviations below the mean on multiple tests or subtests of receptive or expressive language (Leonard, 2014; Spaulding, Plante, & Farinella, 2006).

Assessment of Language Disorders

A complete assessment of language disorder should include a detailed case history, interviews with the individual and his family, ecologically valid standardized and non-standardized measures of communication, and consideration of related areas (e.g., behavioral, medical, educational status). Thus, assessment of language disorder should be a multifaceted process that involves consideration of the aspects of an individual's life that are affected by language. A single individual can collect this data, but the preference is for professionals from different disciplines to work together in the assessment process. According to the World Health Organization, this practice model is called interprofessional practice (IPP). In IPP, professionals work collaboratively in healthcare and education settings to learn about, from, and with each other (World Health Organization [WHO], 2010). When multiple service providers from different professional backgrounds work together, they are better able to provide comprehensive health-care and educational services to individuals and the families they serve.

Infants and Toddlers

Assessment of language disorders in infants and toddlers is primarily related to predicting which children have significant delays in language development that will persist over time. However, the diagnostic accuracy of identifying infants and toddlers with persistent language disorder is relatively poor (Woods, Rieger, Wocadlo, & Gordon, 2014). For this reason, a diagnosis of language disorder is rarely made before 24 months of age.

Test instruments and analyses used for infants and toddlers include items to assess developmental milestones related to communication, motor, cognitive, sensory, and social-emotional skills. An example of a norm-referenced test for measuring general development in multiple domains is the *Battelle Developmental Inventory, Second Edition* (BDI-2) (Newborg, 2005), which can be used to obtain developmental age scores for personal-social skills, adaptive behaviors, communication skills, and cognitive abilities. The *Rossetti Infant-Toddler Language Scale* (Rossetti, 2006) has items measuring gestures, play, language comprehension, and language expression, but it does not compare toddlers to other children their same age. In addition, informal observation and sampling may be used when the infant or toddler may not be able to cooperate with a test procedure because of compliance issues or extreme disability.

Parent report is often the best way to obtain evidence about language and communication skills in infants and toddlers. There are screening and assessment measures such as the *MacArthur-Bates Communicative Development Inventories* (CDI), in which parents of children between 8 and 37 months of age report which words their child understands or uses. The scale divides the words children understand and produce into semantic classes so that examiners can get a sense for the concepts and categories of words that are present or need elaboration. In addition, there is a section that asks parents to report the communicative and symbolic gestures their child has attempted or mastered.

Preschool and School-Age Children

A combination of assessment tools and analyses is needed in order to gain a full understanding of a preschool-age child's receptive and expressive language skills. Some standardized tests assess multiple areas of language to provide a broad view of an individuals' communication system.

Two of these global language measures are the *Clinical Evaluation of Language Fundamentals – 5th Edition* (CLEF-5; Wiig, Semel, & Secord, 2013) and the *Test of Language Development – 4th Edition* which is available for two levels, primary (TOLD-P:4; Hammill & Newcomer, 2008b) and intermediate (TOLD-I:4; Hammill & Newcomer, 2008a). These tests offer composites that represent overall language functioning as well as domain-specific skills. Adding the *Test of Narrative Language-2* (Gillam & Pearson, 2017) to either of these measures would provide examiners with the type of composite scores used in the Tomblin epidemiological studies: overall indices of expressive and receptive language as well as scores representing comprehension and use of vocabulary, grammatical morphology, syntax, and narration. Many other tests are available that assess specific areas of language skill. Examiners should select standardized tests that are reliable and valid in relation to the child being tested, particularly if the child is from a culturally or linguistically diverse background (Friberg, 2010; Paul & Norbury, 2012; Spaulding et al., 2006).

Although standardized tests can give information about an individual's communication functioning relative to a normative population, nonstandardized assessments of language provide a rich description of an individual's communication functioning in multiple contexts. Examples of nonstandardized assessments include language samples, interviews, and dynamic assessment.

Examiners commonly collect a *language sample*, or segment of connected speech, from children in natural environments. Language samples reveal information about the individual's language productivity (total number of words, number of different words, mean length of utterance), language complexity (complex vocabulary and sentence structure), and accuracy (grammatical acceptability and articulatory intelligibility) and may reveal information about the individual's communication that is not readily apparent in standardized measures (Costanza-Smith, 2010). Language samples are collected using a variety of methods. Conversational language samples, in particular, illuminate an individual's ability to comprehend questions and statements within the normal flow of discourse. These can be collected through direct observation by the examiner, or family members may provide videos of an individual's communication with friends, siblings, parents, and others, which can then be analyzed. Language samples are particularly useful for assessing individuals of culturally and linguistically diverse backgrounds (Laing & Kamhi, 2003).

Input from people who interact regularly with a child suspected of having a language disorder further informs the diagnostic process. Parents and other family members provide valuable information regarding birth and development of children; further, they provide critical insight into how the child's communication affects family life, social interactions, and academic functioning. Teachers can also assist, giving information about academic and social functioning within the school setting. As children mature, information from self-reports becomes increasingly important to inform intervention as the individual's priorities need to be considered.

Examiners may also use a process called *dynamic assessment* to improve diagnostic accuracy. Dynamic assessment is a "test-teach-retest" method in which clinicians obtain evidence of a child's ability to perform a language task (such as narration); then observe the child's learning processes as they teach the skill during two teaching sessions; and then retest to determine how much the child learned. This method does not rely on an individual's prior experience, so it is designed to assess an individual's ability to respond to teaching. Observations of the child's ability to modify their responses based on the clinician's input are the key to making correct judgments of language impairment (Pena & Gillam, 2000; Peña, Gillam, & Bedore, 2014).

The assessment of language disorder involves a battery of assessment tools, both formal and informal. Understanding an individual's abilities compared to others of similar age and experience is important for diagnosing language impairment accurately. Collecting information about the individual's functional abilities, strengths, and weaknesses can be even more important for developing intervention plans.

Etiology of Language Disorder

Researchers have hypothesized neurophysiological, biological (genetic), and psychological (cognitive/linguistic) causes of language disorder. As with nearly all of the neurodevelopmental disorders, the precise etiology of language disorder has yet to be determined.

By definition, children with a language disorder present language difficulties in the absence of frank neurological impairments such as brain lesions. However, it is reasonable to assume that there is a connection between neural functions and language ability. To date, relatively little neuroimaging research has been conducted with this population. Some studies have revealed neurological differences between children with language disorders and typically developing children. For example, structural differences have been noted in inferior frontal cortex (Gauger, Lombardino, & Leonard, 1997) and the temporal-parietal junction (Jernigan, Hesselink, Sowell, & Tallal, 1991) in children with a language disorder. In addition, many of these children have been shown to have atypical hemispheric asymmetries in which the perisylvian area (inferior frontal gyrus back to the supramarginal gyrus) in the right hemisphere is larger than the same area in the left hemisphere (Gauger et al., 1997; Plante, Swisher, Vance, & Rapcsak, 1991). This pattern is the opposite of the left lateralization that is seen in most typically developing children. However, fMRI studies (Ellis Weismer, Plante, Jones, & Tomblin, 2005) and functional near-infrared spectroscopy (fNIRS) studies (Gillam, Wan, Gillam, & Hancock, 2015) have not revealed laterality differences between children with and without language disorders as they were engaged in memory or language tasks.

There have been many more electrophysiological studies of children with language disorders using EEG and ERP techniques, primarily because this technology is relatively inexpensive and data can be collected in more naturalistic settings. Results of a number of electrophysiological studies suggest that attention processes for auditory nonspeech and speech stimuli are unusual in children with language disorders (McArthur, Atkinson, & Ellis, 2009; Weber-Fox, Leonard, Hampton, & Tomblin, 2010). Group differences have been detected for grammatical processing (Shafer, Schwartz, Morr, Kessler, & Kurtzberg, 2000; Weber-Fox et al., 2010) but not for semantic processing (Fonteneau & van der Lely, 2008; Weber-Fox et al., 2010). Importantly, it has been shown that ERP responses of children with language disorders become more like their typically achieving peers after participating in a narrative language intervention (Popescu et al., 2009). The EEG and ERP evidence suggests that neural processing differences contribute to language disorder. More importantly, these processes can be altered through language intervention. Clearly, this is a fertile area for future research.

There are genetic contributions to language disorder. A number of studies have shown that children with language disorder have higher percentages of family members with a history of language problems in comparison to control children without a language disorder (Barry, Yasin & Bishop, 2007; Flax et al., 2003). Studies of twins consistently reveal higher concordance rates (both twins presenting language disorder) for monozygotic twin pairs than for dizygotic twin pairs (Bishop, North, & Donlan, 1995; Tomblin & Buckwalter, 1998). Molecular genetic studies have identified potential genetic variants that may contribute to language disorder including FOXP2 (Lai, Fisher, Hurst, Vargha-Khadem, & Monaco, 2001; Tomblin et al., 2009), CNTNAP2 (Vernes et al., 2008), and 6p22 (Rice, Smith, & Gayán, 2009). Rice (2012) has proposed that the start-up mechanisms responsible for expression of genes related to language development may be delayed in children with a language disorder. This hypothesis is consistent with evidence of slower growth trajectories in children with language disorders (McArthur & Bishop, 2004; Rice, Wexler, & Hershberger, 1998; Tomblin et al., 2003).

Psychological hypotheses about the etiology of language disorder can be divided into linguistic (domain-specific) and cognitive (domain-general) factors. Domain-specific explanations of language disorder suggest that the difficulty children with language disorder have is specifically related

to the grammatical system. Most domain-specific accounts of language disorder are related to the notion that some children treat certain linguistic rules as optional rather than obligatory (Marinis & van der Lely, 2007; Friedmann & Novogrodsky, 2007). One early explanation is called the extended optional infinitive hypothesis proposed by Rice, Wexler, & Cleave (1995). Under this account, a language disorder is thought to arise from a specific difficulty in understanding that one must mark tense in a main clause. Inconsistent marking of tense is typical for all children as they acquire language; however, students with language impairment continue to do so long after their typically developing peers (Rice et al., 1998).

Domain-general explanations of language disorder suggest that capacity limitations in cognitive processing interfere with one's ability to learn language. Evidence has shown that children with language disorders demonstrate slower response times during linguistic and nonlinguistic tasks (Kail, 1994; Leonard et al., 2007); however, some have argued that processing inefficiency, rather than slow processing speed, may better explain performance of children with language impairment on these tasks (Gillam, Cowan, & Day, 1995). It is likely that some combination of cognitive factors including limited information processing capacity (e.g., attention, perception, and memory) and slowed processing speed play an important role in language disorder. For example, working memory and processing speed together have been shown to account for 62% of the variance in the composite language scores of adolescents with and without language disorder (Leonard et al., 2007).

Symptom Presentation, Course of the Disorder, and Developmental Changes

Preschool Years

During the preschool years, children should learn a variety of words that represent different word classes (e.g., question words, colors, spatial terms), conjunctions (e.g., coordinating, subordinating), and words that may be used to categorize and describe objects, actions, and events (Anderson, 2011). By 24 months of age, children developing typically are using two-word phrases and have diverse and varied vocabularies, while children with a language disorder may still be pointing at objects they want or using a few single words. Preschoolers who do not learn words easily or who have difficulty expressing a range of semantic relationships such as possession (e.g., mommy's shoe), recurrence (e.g., mommy came back), and location (e.g., mommy in kitchen) may have a language disorder.

Preschoolers with language disorders often produce very few grammatical morphemes (e.g., possessives, plurals) and have difficulties using them to mark tense and agreement (e.g., She walkded home. She my friend). They may also have difficulty understanding and using complex sentences. Preschoolers developing typically generally produce sentences that are grammatically correct (70%) at the age of 3, while preschoolers with language disorder at the same age make significantly more errors (>70%).

School-Age Children

School-age children may continue to make some grammatical errors in their sentences; however, no more than 20% of their sentences should be grammatically incorrect. In addition, the conversational speech of school-age children should contain a variety of complex sentences (at least 20% in a 100 utterance sample). Children with language disorders tend to use fewer complex sentences and tend to make more grammatical errors throughout the school-age years (Gillam & Johnston, 1992).

Another sign of a potential language problem for school-age students includes delays or deficits in phonological awareness (Gillon, 2005). Phonological awareness is the knowledge that individual sounds make up the words in the language. Tasks that measure this awareness include those that requires sound categorization (e.g., which word starts like leg; lamb, fox, ball), blending (e.g., when you combine the sounds /b/, /a/, /l/,

they make the word…?), and segmentation (e.g., the word ball may be broken into what three sounds?). Problems with phonological awareness may make it more difficult for children with language disorder to make the necessary connections between letters and the sounds they represent, making word recognition difficult.

Narration (storytelling) is an important form of discourse during the school-age years. Children developing typically produce narratives that are generally grammatical and contain an adequate number of syntactically diverse and complex sentences (>20%). Their stories contain basic story elements (settings, events, actions, endings) that are organized temporally and logically. Children with language disorder are likely to produce narratives that are disorganized and that contain more ungrammatical sentences and fewer complex sentences (Gillam & Johnston, 1992).

Finally, a hallmark of a language problem during the school-age years is difficulty understanding and using higher-level skills such as inferences (Karasinski & Ellis Weismer, 2010; Laing & Kamhi, 2002). Inferences are assumption that are made about concepts or ideas that were not stated. For example, in the paragraph, "The girl watched while the boy jumped onto the steps. He slipped and fell. She laughed," inferences are required to understand that the child who "slipped and fell" was the boy and that the girl was the one who laughed.

Secondary School

Children with a language disorder at the adolescent stage may demonstrate difficulty with understanding and using figurative language, multiple meaning words, idioms and abstract, or curricular vocabulary. Much of the conversational and written discourse that is encountered during the adolescent years contains morphologically complex words that require knowledge and experience with word structure as well as mastery of sophisticated metalinguistic skills (Nippold & Sun, 2008). Students with language disorders may demonstrate poor comprehension of curricular texts presented orally or in written form identified by an inability to state main ideas; to recall and report facts about persons, dates, and details; and to make logical generalizations and conclusions (Gillam et al. 2008). They may also have difficulties composing book reports, essays, and term papers that require integration of information contained in oral and written discourse.

Sociodemographic Trends in the Identification of Language Disorder

As mentioned earlier, Tomblin et al. (1997) conducted an epidemiological study on the prevalence of language disorders based on a stratified sample from regions of Illinois and Iowa. They estimated an overall prevalence rate of 7.4%. They further estimated a prevalence rate of 6% in girls and 8% in boys, which differs from the 2:1 male to female ratio that had been previously reported in the literature. When race and ethnicity were taken into consideration, variations across groups were found, with Native American children having the highest prevalence rates, followed by African-American children and then Hispanic children. None of the Asian American children were found to have a language disorder in that particular study. Children who spoke another language than English or who heard another language spoken at home were excluded from the study. The differences in prevalence rates could be attributed to culturally and linguistically biased assessments. These differences were further confounded by a socioeconomic factor, namely, racial/ethnic background correlated with parental education, which was also associated with language disorder.

Epidemiological studies reveal cracks in the referral and diagnosis system. Tomblin et al. (1997) found that only 29% of the parents of children identified with a language disorder had been informed by a professional that their child had a speech or language problem. When accounting for severity of the child's disorder, 39% of the parents of the more severely impaired children had prior knowledge of their child's language problems compared to 27% of the less

severely impaired children. Similarly, in a study of 513 at-risk, low SES children, King et al. (2005) found that at least one source (parent, primary care provider, or home visitor) reported initial concerns about 60% of the more severely language impaired children but only 43% of the less severely language impaired children.

Peña, Gillam, Bedore, and Bohman (Peña, Gillam, Bedore, & Bohman, 2011) determined the risk for language disorder in a sample of 1, 100 bilingual prekindergarten children in two states. Using the criteria of performance at or below the 25th percentile on at least one Spanish measure and one English measure, they found that 30.6% of 1029 Latino children were at risk for language disorder. There were no differences in the risk of language disorder for various subgroups of children with differing levels of exposure to English and Spanish when controlling for age. Unlike the Tomblin, Barker, & Hubbs (2007) study, there was a weak association between socioeconomic status and risk for language disorder in this Latino population. When they followed 167 at-risk children over a 2-year period of time, they found that 12% of at-risk bilingual children were language impaired (Gillam, Peña, Bedore, Bohman, & Mendez-Perez, 2013). This is consistent with the King et al. (2005) finding that 10% of their at-risk population had language disorders. The availability of linguistically and culturally appropriate assessments may impact accuracy of prevalence in diverse groups leading to both overrepresentation and underrepresentation of these groups. It appears that a process that involves universal screening for language ability followed by more complete language testing yields a prevalence rate of 10–12% for samples of monolingual and bilingual children.

Associated Impairments

Problems with the acquisition and use of language are prominent characteristics of a number of diagnostic conditions. Language disorder may result from deficits in the growth and development of the brain. Language impairment is also associated with neurocognitive disorders such as traumatic brain injuries or sensory disorders such as hearing impairment. Finally, language disorder can be related to neglect and abuse, behavioral problems, and emotional problems. The most common associated conditions are late talkers (early delayed language), social communication disorder, intellectual disorder, and autism spectrum disorder.

Late Talkers

Children who start speaking very late (fewer than 50 words at 18 months of age) are often referred to as *late talkers*. When these children have age-appropriate language comprehension skills and no family history of language impairment, they are likely to catch up to their peers by the time they reach kindergarten (Rescorla, 2002). They often perform within the normal range on a variety of language measures by the time they reach high school (Rescorla, 2009). Late talkers whose deficits persist into the school-age years are usually identified as having a language disorder.

Social Communication Disorder

An important change in the DSM-5 involves the distinction between disorders that primarily affect semantic and syntactic aspects of language and those that primarily affect the pragmatic aspect of language. Social (pragmatic) communication disorder (SCD) is defined as a primary deficit in the social use of nonverbal and verbal communication as demonstrated by impairments in social communication (greetings, sharing information), in the ability to meet listener needs, and in such skills as taking turns in conversations. These problems limit the ability to develop and maintain social relationships. Many children have symptoms that cross the boundaries between SCD and language disorder. For example, children with impairments of language form (syntax) and content (vocabulary) may have social communication difficulties as well. However, in diagnosing a social communication disorder, the clinician must determine that the child's poor

social language skills do not result primarily from vocabulary or grammar impairments.

Intellectual Disability

An intellectual disability is indicated by severe deficits in intellectual functions (e.g., reasoning and problem solving) and adaptive behaviors (e.g., communication and activities of daily living) that appear during childhood or adolescence. Children with intellectual disabilities often present motor, language, and social delays before 2 years of age. Later in development, they earn scores below 70 on both IQ tests and measures of adaptive functioning. Intellectual disabilities can be caused by genetic conditions such as Down syndrome or fragile X syndrome, maternal infections during pregnancy such as rubella or cytomegalovirus, complications during delivery resulting in anoxia, diseases such as measles and meningitis, or environmental factors such as neglect or severe malnutrition. Children with intellectual disabilities almost always have language impairments related to language content, and many also have impairments in the area of language form (Rakhlin & Grigorenko, 2015). It is common for these children to be significantly delayed in learning words and to use short, simple sentences well into the school-age years. Despite the fact that children with a language disorder do not have deficits in basic intelligence, the nature of the language impairment demonstrated by children in both groups can be quite similar (Laws & Bishop, 2004). The exception is that there is often a bigger discrepancy between comprehension and production abilities in children with a language disorder than in children with an intellectual disability (Polišenská & Kapalková, 2014).

Autism Spectrum Disorder

Children diagnosed with autism spectrum disorder (ASD) must have pervasive and sustained difficulties with reciprocal social communication and social interaction characterized by severe problems with conversation, sharing of interests or emotions, and initiating or responding to social interactions. Children with ASD must also present restricted or repetitive patterns of behavior, interests or activities such as repetitive motor movements (e.g., rocking or rubbing their heads), insistence on sameness, and/or inflexible adherence to routines. These symptoms are present from early childhood, and they interfere with daily activities and the ability to learn through socialization. Language impairment accompanied by a lack of interest in socializing (e.g., lack of eye contact) or unusual social interactions (e.g., not responding to speaker or responding to anything said by saying the same words over and over) may be early signs of ASD. Even though a number of individuals with ASD have intellectual disabilities and/or language disorder, their key symptoms cannot be explained by these other deficits (APA, 2013).

In the past, ASD and language disorder have been viewed as separate and distinct disorders; however, it has become increasingly clear that there are a number of areas in which the two overlap in their language profiles (phenotype), as well as neural processing patterns and genetic contributions (Ellis Weismer, 2014). In terms of language profiles, children with ASD as well as some with language disorder have been shown to demonstrate similar problems in language use or pragmatics. While some have suggested that basic language skills such as phonology, morphology, and syntax remain intact in ASD, recent research has shown that many students with ASD experience more basic language deficits involving all of the domains of language. In fact, as many as 50% of students with ASD never develop functional communication skills (Eigsti, de Marchena, Schuh, & Kelley, 2011).

There are neurological and genetic differences between children with language disorder and children with ASD. While some studies have shown rightward hemispheric asymmetry in children with ASD and children with language disorder, children with language disorder are more likely to have structural differences in inferior frontal cortex (Broca's area) than children with ASD. Genetic studies that have explored potential

overlap between language disorder and ASD have shown that the incidence of ASD is increased for both siblings of children with SLI and ASD; however, there is no clear indication of a common genetic etiology between ASD and language disorder (Rakhlin & Grigorenko, 2015).

Comorbid Disorders

Attention-Deficit/Hyperactivity Disorder

Attention-deficit/hyperactivity disorder is characterized by the presence of developmentally inappropriate levels of hyperactivity, impulsivity, and inattention across multiple contexts and settings that affects 3–5% of the school-age children (National Institutes of Health [NIH] Consensus Development Panel, 2000). Children with ADHD may demonstrate comorbid deficits in language, but it is not uncommon for the presence of ADHD to result in misdiagnosis of language disorder. Generally, children with ADHD without language impairment do not demonstrate significant errors in marking tense, while children with language impairment do (Redmond, 2005). In addition, students with ADHD with no comorbid deficits in language have been shown to perform similarly to typically developing peers in producing organized stories that contained critical story elements (events, actions, endings), while students with language impairments do not (Luo & Timler, 2008). In a recent study, Redmond, Thompson, and Goldstein (2011) examined the extent to which tense marking, nonword repetition, sentence recall, and narrative proficiency accurately characterized language impairment in 60 7–8-year-old children. Findings revealed that students with ADHD performed similarly to their typical peers on measures of tense marking, nonword repetition, and sentence recall, while the students with language impairment did not.

Specific Learning Disorder

Specific learning disorder (more commonly referred to as a learning disability) is characterized by persistent difficulties learning academic skills. These children's skills in the areas of word decoding, reading comprehension, spelling, writing, number facts, and/or mathematical reasoning are well below the expectations for their chronological age. These learning disorders are "specific" because, like language disorder, they cannot be explained by intellectual disabilities, visual or hearing problems, other mental or neurological disorders, or poor instruction.

Treatment for Children with Language Disorders

Language intervention for children with language disorder involves techniques designed to remediate or modify the factors that contribute to language disorder or teaching compensatory strategies to reduce the impact of a language disorder on academic, social, and/or vocational functioning. Clinicians design treatment programs that target deficiencies in language form, content, and use for preschoolers (ages 3–5), elementary students (ages 5–10), and adolescents (11–21). In the next sections, we will discuss language targets (form, content, and use) as well as methods and techniques used to remediate, modify, or help children compensate for factors that contribute to difficulties in these language domains.

Intervention for Preschoolers

Targets Intervention targeting language form in preschoolers typically focuses on improving morphological abilities such as the use of copula *be* (am, is, are, was, were), auxiliary *be* (am, is, are, was, were), or auxiliary *do* (do, does, did). Other forms that might be targeted in intervention sessions include the use of possessive -s, third person present -s, and irregular and regular past tense verbs. Clinicians may work with preschoolers to increase the length and complexity of the sentences they use and to use more diverse types of sentences such as statements, commands, and interrogatives.

Intervention for preschoolers may also focus on the content of language. For example, clinicians may work with children to increase the size of their vocabulary (e.g., basic concepts, colors),

their use of various semantic relationships (e.g., agent-action, agent-action-object), and their understanding and production of verbs, pronouns, and conjunctions.

Language use or pragmatic goals may also be a part of language intervention programs for preschool children. Intervention for language use might include teaching children to be more flexible in their use of language in different contexts, to use their imagination, and/or to improve their abilities to participate and contribute to conversations they have with parents, teachers, and their peers. Clinicians may help preschoolers develop their conversational skills by improving their abilities in initiating and maintaining conversational topics, in taking turns, and in knowing when and how to change topics. In addition, preschoolers with language disorders may also need to learn how to let others know when they do not understand something and to clarify information they provide during conversations that may be misunderstood by others (Brinton & Fujiki, 1995).

Clinicians may also work with preschoolers on narrative comprehension and production. Preschoolers with narrative language difficulties may work on their abilities to answer basic questions about stories such as "who did what to whom?" Clinicians may work with preschoolers to relate past experiences, to describe ongoing activities or routinized events, or to recount or generate fictional stories. All aspects of language form and content necessary for preschoolers to understand and produce narratives would be integrated into this kind of intervention program.

Preschoolers with language disorders may develop problems in literacy, so intervention might also target emergent literacy skills such as print awareness, book awareness, and letter-sound correspondence or activities designed to foster phonological awareness. Phonological awareness lessons may involve teaching preschoolers to blend and segment spoken words, to delete whole words from compound words, and to delete syllables and phonemes from spoken words, phrases, or sentences (Goldstein & Olszewski, 2015).

Methods and Techniques for Improving Language Form, Content, and Use for Preschoolers There are three basic approaches for providing intervention services to young children with language impairments. These include clinician-centered, child-centered, and hybrid approaches (Fey, Long, & Finestack, 2003). Clinician-centered approaches include very structured techniques such as engaging children in drill and drill-play activities. Child-centered techniques include indirect stimulation approaches. Hybrid approaches incorporate both clinician-centered and child-centered techniques.

Clinicians may use various indirect language facilitation techniques and contingent responses within each of these general approaches to model the forms, content, and pragmatic skills the child needs to learn. Indirect language facilitation techniques may include *demonstrations, expansions, expatiations, and vertical structures* (see Cleave, Becker, Curran, Van Horne, & Fey, 2015 for a meta-analysis of the efficacy of these techniques). Demonstrations are the repeated but variable use of linguistic patterns during ongoing play activities, such as when playing with a toy farm set. For example, to highlight the use of past tense -ed, a clinician might say, "The pig trotted into the barn. The cow trotted into the barn. The goat trotted into the barn. They all trotted into the barn."

Expansions are contingent verbal responses that increase the length or complexity of an utterance produced by the child during the play interaction. For example, a child might say, "Puppy" to which the clinician might say, "Yes, that is a puppy." In this example, the utterance "Yes, that is a puppy" is used contingently by the SLP because it incorporates what the child said (puppy), and it is an expansion because it incorporates the child's utterance into a longer more complex utterance from one (puppy) to five (yes, it is a puppy).

Expatiations are contingent verbal responses that add new but relevant information to the child's utterance. For example, a child might say "goat" to which the clinician may respond "That is a stinky goat." The utterance provided by the clinician is contingent because it includes what the child offered (goat), but it is also an expatiation because it adds new information (There is a goat AND it is stinky).

Vertical structures are language facilitation techniques that involve asking children questions to elicit at least two responses from them. These responses are then combined by the clinician into

one longer, more complex utterance. For example, consider the following interaction targeting the causal connective "because":

Clinician:	The pig got a new hat. How do you think he feels?
Preschooler:	Happy.
Clinician:	Why does he feel happy?
Preschooler:	Because he got a hat.
Clinician:	The pig is happy because he got a new hat.

When using vertical structuring as a language facilitation technique, it is important to remember that both utterances that are elicited from the preschooler must be combined into a complete sentence (happy, because he got a new hat). Simple repetitions of each of the utterances independently do not constitute a vertical structure.

Language Intervention During the Elementary School Years (Ages 5–10)

Targets Language intervention during the elementary school years (ages 5–10) targets similar language skills as those of the preschool years (e.g., morphology, sentence structure, vocabulary, pragmatics, and literacy) but may incorporate more advanced forms using curriculum-based instructional materials (e.g., books, classroom conversation). For example, elementary students may need morphological intervention to help them understand and use advanced morphological forms (e.g., prophet/prophetic), prefixes, and suffixes (e.g., dis-, inter-, -ic, -tion) and to be more aware of grammatical errors they may be making in spoken and written language. Further, students may need to learn and practice certain sentence structures such as those that contain dependent clauses (e.g., prepositional phrases, adverbial clauses) so they may better understand and produce more complex sentence structures.

Language content or vocabulary instruction may target understanding and use of basic word knowledge, abstract or figurative language, as well as literate language such as coordinating and subordinating conjunctions, adverbs, metalinguistic and metacognitive verbs, and elaborated noun phrases. In addition to teaching students new words, clinicians may also assist them in learning strategies, so they may learn new words on their own.

Intervention for language use (pragmatics) during the elementary school years involves helping students to learn the discourse management skills they need to participate in conversations and to repair conversational breakdowns. Other conversational skills that may be part of an intervention program for elementary students with language disorder may include teaching students to request clarification when they do not understand and to express their ideas in polite or persuasive ways in a variety of contexts.

Students may also need instruction to help them understand and use narrative and expository discourse skills. Fictional narratives contain characters whose goal-motivated actions become part of a story. Students with language disorders may need instruction to learn how to identify the parts of a story (character, setting, initiating event, action, consequence), so they may better organize their own stories and recall stories they have heard or read. Instruction may also focus on assisting students in telling stories that contain multiple embedded or associated episodes using coherent and organized linguistic form and content.

Expository discourse such as comparison, cause and effect, or problem-solution becomes important in academic settings during the elementary school years (Gillam, Fargo, & St. Clair Robertson, 2009). Clinicians may provide students with language disorder instruction on how to identify features of the different types of expository structures. For example, to identify comparison texts, students would be taught signal words such as *different from*, *same as*, *similar to*, *instead of*, and *although.* Students would then be taught how to use these words *to* compare how two or more events, topics, concepts, theories, problems, or objects compare.

Clinicians may focus on helping elementary students with language disorder to learn the underlying language skills they need to become proficient readers. For example, students in the early grades learn the difference between long and short vowels and how to blend sounds into

words while reading. Clinicians may design lessons to strengthen their phonological processing skills. Higher-level phonemic awareness tasks such as deletion (e.g., deleting sounds from clusters) and practice with multisyllabic words (e.g., intellectual) might be part of such a program.

Methods and Techniques for Improving Language Form, Content, and Use for School-Age Students Several treatment procedures including the clinician-centered, client-centered, and hybrid approaches used with preschoolers are appropriate to improve language form, content, and use with school-age children. Another popular approach to language intervention during the school-age years, called literature-based language intervention, incorporates aspects of modeling, practice, role-play, and discussion.

Literature-based language intervention utilizes children's literature as a context for teaching specific form (morphology, syntax), content (semantics/vocabulary), use (narrative, expository, and conversational discourse), and literacy targets (phonological awareness) that students may work on to be successful in multiple settings. Literature-based activities typically include clinician-led discussions about concepts and ideas that are contained in the books. Clinicians may read and reread a story, pointing out specific aspects in the story that are relevant to the goals the student is working on. For example, students may be working on narrative discourse comprehension and retelling. In this case, the clinician might read a story, highlight the story elements that are contained in the story (character, setting, events), and then ask students to answer questions about the story elements. The clinician may reread the story and then ask students to retell the story using the pictures in the book and then, without them, to foster recall and memory-specific details.

If language form is a target, the clinician may reread the book while highlighting the form that is the focus of instruction at that point in time, for example, causal words such as "because" or "so." The clinician may use vertical structuring to facilitate student's use of complex sentences containing the words such as in the following example using the book *Tacky the Penguin*:

Clinician: How did the penguins in the book feel about the hunters?
Student: The penguins were afraid of the hunters.
Clinician: Why were they afraid?
Student: Because the hunters might get them.
Clinician: The penguins were afraid because the hunters might get them.

Similarly, vocabulary is a frequent target of literature-based language intervention. Literature-based vocabulary instruction is most effective when words are embedded in stories that are read repeatedly, when the meaning of words are explicitly taught within the story context, and when students are given opportunities to use the target vocabulary during their own discussions and retellings about the text (Biemiller & Boote, 2006).

In one recent study, Gillam, Gillam, and Reese (Gillam, Gillam, & Reece, 2012) found that literature-based language intervention that incorporated these important aspects of instruction resulted in larger narrative and vocabulary gains than more clinician-directed, drill-based language intervention procedures.

Language Intervention During the Adolescent Years (Ages 11–21)

Targets *Adolescent language intervention* often focuses on targeting skills to support students, so they may meet the increasing linguistic demands of middle and high school. This is important if they are going to be ready to enter the workforce or to be prepared for college entry should they choose that path. Intervention programs for adolescents will target higher-level language skills they need to understand curricular texts such as the use of inferencing and the comprehension of figurative language and advanced vocabulary, including words they need to understand to access their curricular materials. In addition, clinicians will assist students in learning the language skills they need to analyze and construct well-supported arguments.

Unfortunately, not all activities are designed to remediate or modify a language disorder particularly in the adolescent years. Very often, clinicians must teach compensatory strategies to students at this stage, so they are able to circumvent some of the underlying problems that may not be remediable. Strategies include teaching students how to use graphic organizers, checklists, and organizational techniques in order to help them recall and use information to answer questions, recall information, and construct spoken and written arguments. Strengthening students' metalinguistic and metacognitive skills will also be an important part of intervention efforts during the adolescent years. Students may be taught specific strategies they may use to be able to monitor their own performance during reading, writing, and even in social contexts. In addition to teaching strategies and self-monitoring skills, clinicians may need to put certain modifications in place to help students to be more successful in the classroom such as preferential seating and making recorded lectures available to them.

Methods and Techniques for Improving Language Form, Content, and Use for Adolescents Many of the same methods and techniques used for younger school-age students may also be effective for adolescents and include direct instruction during literature-based activities; however, because adolescents are more cognitively sophisticated, they may take part in more metacognitive activities that involve encouraging them to think about what they know and how they learn best. Role-playing becomes an important intervention context at this stage and may involve presenting students with situations where they must generate solutions that target form and content or use goals that are important for them. For example, students may be asked to provide a solution to a scenario that takes into consideration how characters feel and does not compromise the relationship between the two characters. Further, students may be asked to explain "why" they feel their solutions are appropriate in different situations. In particular, adolescents must be able to generate long-term solutions to problems and discuss why short-term solutions may not be sufficient. Any number of language targets, strategies, and self-monitoring skills may be taught during role-playing with adolescents and is a popular method for teaching at this stage.

Contexts of Intervention

In years past, removing one or more students from the classroom has been the traditional method for providing intervention to students of all ages. More recently, in light of the World Health Organization's call for interprofessional practice in health-care and educational settings, clinicians and educators have become more comfortable working together, sometimes in regular classroom, to provide services to children with language disorders. This approach to language intervention makes it possible to integrate language-learning goals (complex sentence usage, narrative abilities, phonological awareness, vocabulary, expository texts, learning strategies, etc.) into aspects of the academic curriculum. Clinicians and classroom teachers may work together to plan lessons and will either carry them out together with the whole class or one of them may lead the class while the other facilitates student participation. For example, a clinician might conduct lessons on creating narratives with the entire class all at once (Gillam, Olszewski, Fargo, & Gillam, 2014), while the classroom teacher works with small groups of students in the classroom by helping them ask or answer relevant questions.

Another possibility is to arrange the classroom into small "centers" that allow for groups of children to rotate through them to practice various phonological awareness skills (van Kleeck, Gillam, & McFadden, 1998) or even to develop new vocabulary (Gillam, 2007). For example, students may participate in a listening center, where they hear definitions of new vocabulary prerecorded on a digital recorder that are relative to a curricular topic, such as volcanos. In the next center, they may watch a short film about volcanos. In the last center, students may participate in discussions with the clinician and asked to provide definitions and explanations about the concepts and ideas they are learning. Any number of language targets may be taught in this format. The overwhelming findings

suggest that speech-language pathology services may be effectively provided in regular classroom contexts (Cirrin et al., 2010; Hadley, Simmerman, Long, & Luna, 2000).

Prognosis and Outcomes of Children with Language Disorder

Unfortunately, symptoms related to language disorder tend to persist. There have been a number of longitudinal studies that have followed children diagnosed with language disorders as preschoolers or kindergartners into their teenage years. Approximately 70% of these children continued to demonstrate language impairments as teenagers (Beitchman & Wilson 1996; Nippold, Mansfield, Billow, & Tomblin, 2009). The results of these studies suggest that teenagers with a history of language disorders are highly likely to present with cognitive, linguistic, social, and academic problems (Johnson et al., 2010; Lindsay, Dockrell, Nippold, & Fujiki, 2012; Nippold et al., 2009; Tomblin & Nippold, 2014). Studies of adults with a history of language disorder during childhood indicate that they often have persisting difficulties with measures of language and literacy, poor memory skills, lower levels of employment, and fewer close friendships than individuals with no history of language disorder (Clegg, Hollis, Mawhood, & Rutter, 2005; Johnson, Beitchman, & Brownlie, 2010). However, these individuals are likely to perceive their quality of life in much the same way as their non-impaired peers (Johnson et al., 2010; Records, Tomblin, & Freesse, 1992). Individuals who had less severe impairments during childhood tend to have the best language outcomes at high school and into adulthood. Specifically, in a well-designed longitudinal study, language ability during childhood was a significant predictor of educational attainment and occupational SES during adulthood, especially when it was combined with teacher ratings of behavior, family SES, and reading ability (Johnson et al., 2010). It is clear that there is a strong need to develop more effective interventions for children with language disorders and for teachers, parents, and other professionals who work with these children to advocate for educational and vocational programs that will promote better lifelong outcomes.

Conclusion

Language disorder can be diagnosed (1) when children's receptive and/or expressive language skills are significantly poorer than the language that is typically expected for same-age peers, (2) when his or her language difficulties interfere with the ability to participate in and learn from everyday interactions with adults and other children, and (3) when the language deficits cannot be attributed to any other disability. Neurophysiological, biological (genetic), and psychological (cognitive/linguistic) factors contribute to language disorder.

Very young children with a language disorder may not combine words to express relationships until well after 2 years of age. During the preschool years, children with language disorders may learn new words more slowly and often make more grammatical errors than their typically developing peers. During the elementary school years, children with language disorders have difficulty learning and using words that are important for functioning in their classrooms; their conversational language may contain an unusually large percentage of ungrammatical utterances; and they may experience difficulties understanding and creating coherent narratives. Older school-age children with language disorders do not comprehend and use morphologically complex words like *summarization* and *predictive* that appear in the curricular materials they encounter in school, and they have difficulty understanding the expository texts they read for their courses.

Language disorder can be diagnosed reliably in the late preschool and early school-age years. Clinicians assess language abilities with the use of standardized tests and informal assessment procedures such as language sample analysis, observations of children as they are conversing with others, and interviews with parents and teachers. Clinicians can conduct assessment independently,

or they can employ interprofessional approaches to assessment in which professionals from different disciplines conduct evaluations.

Three basic models of language intervention services were described: child-centered, clinician-centered, and hybrid, as were facilitative techniques such as self-talk, parallel talk, focused stimulation, expansion, and vertical structures. Many clinicians use book discussions as the primary context for language intervention with school-age children. Clinicians who work in public school settings sometimes conduct language intervention in the regular classroom. This approach to language intervention is helpful when clinicians want to integrate the children's language-learning goals (complex sentence usage, narrative abilities, phonological awareness, vocabulary, expository texts, learning strategies, etc.) with the expectations of the academic curriculum.

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Specific Learning Disorder

4

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Introduction

Controversies related to the definition of specific learning disability and the methods used to diagnosis this condition have long been a foremost concern in assessment-related fields, such as school psychology and neuropsychology (e.g., Decker, Hale, & Flanagan, 2013; Reynolds & Shaywitz, 2009), as well as in the medical and legal fields (e.g., Colker, Shaywitz, Shaywitz, & Simon, 2013). The publication of new criteria for diagnosing a specific learning disorder in the 5th edition of the *Diagnostic and Statistical Manual* (DSM-5; American Psychiatric Association [APA] 2013) has fueled such controversies (e.g., http://www.landmarkoutreach.org/about/newsworthy/dyslexia-dropped-dsm-5). This chapter begins with a brief history of learning disabilities, including the diagnostic criteria for learning disorders in the five editions of the DSM. Next, etiology, associated impairments and comorbidities, course and developmental changes, prognosis, outcomes, and treatment are highlighted. This chapter concludes with a brief discussion of the most salient vagaries inherent in the latest diagnostic criteria for specific learning disorder in the DSM-5, some of which will be debated for some time to come. [Note that "SLD" is used in this chapter to refer to "specific learning disorder" as defined in DSM-5, rather than "specific learning disability" as defined in the Individuals with Disabilities Education Improvement Act (IDEIA, 2004).]

Brief History of Learning Disabilities

Between the years of 1800–1920, the field of learning disabilities began to emerge as the result of developments in the fields of neurology and reading disabilities. As Franz Joseph Gall explored relationships between head injury and movement and sensation, morality, and intellectual qualities during the early 1800s, groundwork was set for localization of brain function, and Broca's area was identified as responsible for speech production (Hallahan & Mercer, 2001). Toward the latter part of the nineteenth century, Sir William Broadbent identified the relationship between the loss of reading abilities and the loss of the power to name (e.g., printed letters, familiar objects) following neurological symptoms. Kussmaul's work described what he

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termed "word blindness," which is possibly the first iteration of a specific learning disability as a disorder separate from other potential problems (Hallahan & Mercer, 2001). Similar case studies describing reading disabilities were reported by Hinshelwood and Morgan in the late 1800s and early 1900s. Hinshelwood also studied congenital and acquired reading disabilities and reported on six cases appearing within a family over two generations. Through these studies, he hypothesized that the primary deficit in these children was visual memory for words and letters, what might now be defined as an orthographic processing deficit (Hallahan & Mercer, 2001; see also Berninger, Abbott, Nagy, & Carlisle, 2010).

Interested in the earlier research of the aforementioned European scientists, several American researchers including Orton, Fernald, Monroe, and Kirk continued the study of learning disabilities ranging from language and reading to perceptual-motor and attention disabilities. Orton determined that many students with reading disabilities had average to above-average scores on the Stanford-Binet IQ test, as Hinshelwood had also suspected. Orton's work significantly contributed to interventions for reading disabilities and emphasized a multisensory approach (Hinshelwood, 1902; Orton, 1925).

A former research associate for Orton, Marion Monroe experimented with his methods for remediation as well as the methods of Fernald and Keller. Developing diagnostic tests and using results to inform instruction, she developed a synthetic phonetic approach and compared it to other methods (Hallahan & Mercer, 2001). Monroe was the first to suggest that reading disabilities could be identified by examining the discrepancy between actual achievement and expected achievement, the latter of which was an average of the child's chronological, mental, and arithmetic grade (Monroe, 1932). Monroe also emphasized error analysis in order to guide instruction and laid the foundation for the discrepancy approach and for the response-to-intervention approach to specific learning disability identification (Hallahan & Mercer, 2001; Hallahan & Mock, 2003).

The First Definition of Learning Disability

Working alongside Monroe as an instructor while earning his doctorate, Samuel Kirk studied interventions for reading disabilities and provided what is considered to be the first definition of the term "learning disability." His definition linked brain dysfunction to behavioral manifestation of academic problems. According to Kirk, "A learning disability refers to a retardation, disorder, or delayed development in one or more of the processes of speech, language, reading, writing, arithmetic, or other school subject resulting from a psychological handicap caused by possible cerebral dysfunction and/or emotional or behavioral disturbances. It is not the result of mental retardation, sensory deprivation, or cultural and instructional factors" (Kirk, 1962, p. 263).

During the 1970s, Kirk continued to develop assessment techniques to identify learning disabilities in children. His contribution to the field of learning disabilities, including the *Illinois Test of Psycholinguistic Abilities* (Kirk, McCarthy, & Kirk, 1967), and his definition of the disorder provided the foundation for later researchers to explicate the relationship between cognitive functions and academic problems. Integrating Monroe's discrepancy-based definition of learning disabilities and Kirk's brain-based definition, Barbara Bateman proposed an alternative definition in 1965:

"Children who have learning disorders are those who manifest an educationally significant discrepancy between their estimated potential and actual level of performance related to basic disorders in the learning process, which may or may not be accompanied by demonstrable central nervous system dysfunction, and which are not secondary to generalized mental retardation, educational or cultural deprivation, severe emotional disturbance, or sensory loss" (Bateman, 1965, p. 220). Since Bateman's reintroduction of the concept of discrepancy from cognitive potential, discrepancy has remained a central feature of most definitions, including the *response-to-intervention* approach, and has at times been a controversial feature of later definitions (see Flanagan & Alfonso, 2011, for a comprehensive review).

Contemporaneously, the federal government began its own efforts to provide a definition through the creation of two task forces; however, these task forces failed to provide a unified definition of learning disabilities. Task Force I provided a definition on "minimal brain dysfunction," while Task Force II defined learning disabilities. The disparate definitions reflected the fact that Task Force I was comprised primarily of medical professionals, while Task Force II was comprised of educators.

Shortly thereafter, in the late 1960s, the US Office of Education (USOE) formed the *National Advisory Committee on Handicapped Children (NACHC)* to provide a definition that could serve as the basis for legislation and funding. As Kirk chaired this committee, the resulting definition was highly similar to the definition he provided in 1962, and much of the language used in this definition is present in current definitions (e.g., IDEIA, 2004). The NACHC definition of 1968 emphasized a "disorder in one or more of the basic psychological processes" manifesting as disorders of "thinking, talking, reading, writing, spelling, or arithmetic" and excluded deficits related to visual, hearing, or motor handicaps, to mental retardation, emotional disturbance, or to "environmental disadvantage." In 1969, the Children with Specific Learning Disabilities Act was passed, which was included in the Education of the Handicapped Act of 1970 (PL 91–230), and provided for programs to serve children who were eligible (Hallahan & Mercer, 2001; Hallahan & Mock, 2003).

In 1975, McCarthy attempted to operationalize the definition of the NACHC by providing criteria. The first of these criteria included an IQ greater than 85 as well as sufficient opportunities for learning. The second of McCarthy's criteria included a "significant discrepancy" between academic aptitude and academic achievement and further suggested in the third criterion that the discrepancy was sizable enough to warrant specialized intervention (Tylenda, Hooper, & Barrett, 1987). McCarthy's inclusion of an IQ above 85 is closely aligned with some current research-based definitions of specific learning disabilities (e.g., Berninger, 2011; Flanagan, Ortiz, & Alfonso, 2013; Flanagan, Ortiz, Alfonso, & Mascolo, 2002; Hale & Fiorello, 2004; Kavale, Kauffman, Bachmeier, & LeFever, 2008; McDonough & Flanagan, 2016; Ortiz, Flanagan, & Alfonso, 2015).

Toward Consensus on the Definition of Learning Disability and Methodology

From about 197 to 51985, there was relative stability in the field as it moved toward consensus on the definition of learning disabilities as well as methods of identifying the condition. Applied research was abundant at this time, much of which was funded by the USOE. This research led to empirically validated educational procedures for students with learning disabilities (Hallahan & Mercer, 2001).

Following the adoption of a federal definition of learning disabilities, organizations and researchers focused their efforts on developing and testing intervention programs for individuals with learning disabilities, and the definition of learning disabilities remained relatively stable (Hallahan & Mercer, 2001). Applied research funded by the USOE established a body of empirically validated methods to intervene with children with specific learning disabilities. The definition of "specific learning disability" was further solidified by the passing of the Education for All Handicapped Children Act (PL 94–142), which essentially adopted the 1968 definition proposed by Samuel Kirk and the NACHC. This definition has survived conceptually in the federal legislation in its entirety with only minor text revisions (Herr & Batemen, 2003; Sotelo-Dynega, Flanagan, & Alfonso, 2011).

Public Law 94–142 provided legitimacy and, equally as important, funding, for direct services for children with specific learning disabilities; in addition, the legislation set forth regulations as a way to deal with the absence of prescriptive guidance with regard to the methods used to identify learning disabilities (Sotelo-Dynega et al., 2011). These regulations continued to stipulate the presence of an ability-achievement discrepancy until the most

recent reauthorization of this act – the Individuals with Disabilities Education Improvement Act (IDEIA, 2004) – and its attendant regulations (Federal Register, 2006), the latter of which stated that ability-achievement discrepancy may be used as a method of identifying children with specific learning disabilities, but could not be mandated. Shortly after PL 94–142 was enacted, the DSM included discrepancy criteria for learning disorders.

Disorders of Learning in the DSM

Even in the earliest versions of the DSM, there was some recognition of learning difficulties as a distinct problem facing some individuals. The first edition of DSM (APA, 1952) briefly referred to a "learning disturbance" under the category of "special symptom reactions," as well as to "specific learning defects," including alexia and agraphia. The definition included a reference to "word blindness" as initially described by Kussmaul nearly 60 years earlier (Loriaux, 2010). Coding of the latter diagnosis was altered if known to be related to organic brain dysfunction. Despite the work of many contemporary American researchers in the field, the first edition of the DSM's definition of learning problems was limited in detail and scope.

The second edition of the DSM was published several years following Samuel Kirk's initial definition of specific learning disabilities, and was again listed under a main category of "special symptoms" as a "specific learning disturbance" (APA, 1968).The definition or specification of symptom presentation in the DSM-II was even more limited than those provided in the first edition, despite proliferation of research and published work in the field of learning disabilities.

However, the third edition of the DSM devoted significantly more attention to the issue of defining learning disabilities (APA, 1980). The DSM-III definition included six disorders, namely, developmental reading disorder, developmental arithmetic disorder, language disorder, expressive and receptive language disorders, articulation disorder, and mixed developmental disorder. This definition also included a statement that while mutually exclusive from "pervasive developmental disorders," a specific learning disorder in reading or arithmetic could coexist with a diagnosis of mental retardation (p. 94); this apparent contradiction served to confuse the issue of defining learning disabilities.

The DSM-III provided definitions and criteria for developmental reading disorder and developmental arithmetic disorder. The essential feature of these disorders was a "significant impairment" in the academic skill not accounted for by mental age, chronological age, or schooling. Diagnostic criteria for each of the disorders required a significant discrepancy between intellectual functioning as measured by an individually administered IQ test and performance on an academic achievement test measuring relevant domains. The DSM-III also recommended a 1- or 2-year delay in measured reading skills for children ages 8–13 years. The discrepancy between a full-scale IQ and academic achievement remained central to the definition and identification of specific learning disability.

As compared with the diagnosis of "academic skills disorders" in DSM-III, the DSM-IV (APA, 2000) presented the term "learning disorders" and three specific diagnoses within this overarching category: reading disorder, mathematics disorder, and disorder of written expression. In addition, learning disorder not otherwise specified was included in the DSM-IV and encompassed academic problems that did not meet criteria for one of the aforementioned specific academic disorders; this diagnostic label was intended to be applied to individuals presenting problems across reading, math, and/or written expression or who did not meet the threshold set forth by the term "substantially below" (i.e., achievement substantially below IQ) provided that the observed level of academic performance significantly interfered with academic achievement. The DSM-IV also moved these disorders from Axis II to Axis I.

The DSM-IV sought to improve the definition of learning disabilities over the previous version. The definition again relied on a discrepancy approach, stating "Learning Disorders are diagnosed when the individual's achievement on individually administered, standardized tests in

reading, mathematics, or written expression is substantially below that expected for age, schooling, and level of intelligence" (DSM-IV-Text Revision [TR], 2000, p. 49). The DSM-IV went on to define further "substantially below" as a difference of at least two standard deviations between achievement and measured intelligence. The DSM-IV provided separate definitions for reading, mathematics, and writing disorders, and the second criteria required that the "disturbance" identified in the first criterion (i.e., achievement as measured by individually administered standardized tests is substantially below expected) significantly interfered with academic achievement or with completing activities requiring that skill. And finally, DSM-IV criteria required an impairment above and beyond a sensory deficit if one was present.

As in the DSM-III, the DSM-IV stipulated that the academic deficits were observed on "individually administered standardized tests," thereby assuring a formal evaluation of academic skills in addition to a measure of intelligence. The DSM-IV also referenced the possibility of coexisting specific cognitive processing deficits in an area such as memory, visual-spatial processing, or the like, but did not require that one be present.

In terms of differential diagnosis, the DSM-IV required that learning disorders are differentiated from "normal variations in academic attainment,… lack of opportunity, poor teaching, or cultural factors" (APA, 2000, p. 51) and should be beyond what one would expect associated with a sensory impairment, if present. According to DSM-IV, as in DSM-III, a learning disorder was also possible in addition to a diagnosis of mental retardation, provided the academic achievement was significantly below estimated intellectual functioning. The recent publication of the DSM-5 (APA, 2013), however, represents a significant departure in criteria for learning disorders as compared to previous editions.

DSM-5 Diagnostic Criteria

The DSM-5 resulted in considerable changes to the diagnostic criteria for SLD, and of the most significant changes, reading, mathematics, and writing disorders were combined into one overarching diagnostic category. Tannock (2013) described the rationale behind this change as reflecting the workgroup's conceptualization of SLD as a single disorder that renders learning as very difficult and effortful. Furthermore, whereas subtypes describe mutually exclusive disorders, specifiers allow for concurrent diagnoses of impairment in multiple academic domains. Therefore, the criteria were also altered to include symptoms of reading, writing, and mathematics impairment. Criterion A of the DSM-5 description of SLD is "difficulties learning and using academic skills as indicated by the presence of at least one of the following symptoms that have persisted for at least 6 months, despite the provision of interventions that target those difficulties" (APA, 2013, p. 66). Symptoms include inaccurate or slow and effortful word reading; difficulty understanding the meaning of what is read; difficulties with spelling; difficulties with written expression; difficulties mastering number sense, number facts, or calculation; or difficulties with mathematical reasoning.

Criterion B requires that academic skills be "substantially and quantifiably" below what would be expected based on chronological age and cause significant interference with academic or occupational performance or with activities of daily living. Criterion B also requires that the determination regarding academic skills be based on individually administered standardized achievement measures and "comprehensive clinical assessment." For individuals over age 17 years, a documented history of learning impairment may be substituted for the standardized assessment.

Criterion C states that "the learning difficulties begin during school-age years, but may not become fully manifest until the demands for those affected academic skills exceed the individuals limited capacities" (APA, 2013, p. 67), thereby allowing for situations in which older individuals can be newly diagnosed with an SLD. The DSM-5 provides examples of situations of such demands including timed tests, reading or writing lengthy complex reports for a tight deadline, and excessively heavy academic loads.

Finally, Criterion D remains similar to previous versions of the DSM and stipulates that the

learning difficulties are not better accounted for by intellectual disabilities, uncorrected visual or auditory acuity, other mental or neurological disorders, psychosocial adversity, lack of proficiency in the language of academic instruction, or inadequate educational instruction. Criterion D contains the only reference to cognitive ability in its provision that the academic deficits are not secondary to intellectual disability. A note below the four listed criteria states that they are to be met "based on a clinical synthesis of the individual's history" including developmental, medical, family, and educational histories, "school reports, and psychoeducational assessment" (APA, 2013, p. 67).

The unification of three separate learning disorders into one diagnostic category necessitated the addition of specifiers in order to identify the area(s) of academic weakness; a coding note indicates that when more than one domain is impaired, more than one specifier can and should be assigned. The coding note also directs the clinician to document the subskills that are impaired as part of the diagnosis. The specifier "with impairment in reading" is appended to the SLD diagnosis when the individual demonstrates significant impairment in one or more of the subskills including word reading accuracy, reading rate or fluency, and/or reading comprehension. Dyslexia may be used as an alternative term referring to problems with word reading fluency or word reading accuracy, decoding, and spelling, and, if used, impaired subskills should also be identified. An impairment in writing skills is assigned to the specifier "with impairment in written expression" and refers to those children with impaired spelling accuracy, grammar and punctuation accuracy, and/or clarity or organization of written expression. The specifier "with impairment in mathematics" is used for individuals who demonstrate significantly below average skills in number sense, memorization of arithmetic facts, accurate or fluent calculation, and/or accurate math reasoning. The term "dyscalculia" can be used to refer to difficulties processing numerical information, learning arithmetic facts, and performing accurate or fluent calculations. As when using the term "dyslexia," the use of the term "dyscalculia" also requires the identification of any additional impaired subskills.

In addition to specifying the domain of impairment, the degree of severity should also be indicated in the diagnosis. The range of severity includes "mild," "moderate," and "severe." An SLD of "mild" severity applies to an individual with some difficulties in one or two academic domains but is able to function well when provided with appropriate accommodations or support services. The "moderate" specifier is used when there are marked difficulties in one or more academic domains, and the individual is not likely "to become proficient without some intervals of intensive and specialized teaching during the school years" (APA, 2013, p. 68). Accommodations or supportive services may be needed in school, in the workplace, or at home in order for activities involving the academic skills to be completed accurately and efficiently. A "severe" severity specifier is used to describe impairments in individuals who are unlikely to learn those skills without "ongoing intensive individualized and specialized teaching for most of the school years" (APA, 2013, p. 68). Even with accommodations, an individual with a severe SLD may not be able to perform academic tasks with efficiency.

Etiology

There is consensus among experts in the field that there is no known single cause but rather numerous factors that contribute to the development of learning disabilities. According to the DSM-5, SLD is a neurodevelopmental disorder with a biological basis resulting in cognitive abnormalities; academic skill deficits are considered the behavioral sign or manifestation of the disorder. Genetic, epigenetic, and environmental factors interact to impair the brain's ability to perceive or process information efficiently or accurately (APA, 2013). Table 4.1 provides a summary of the etiology of SLD along with information on impairments that are associated with this condition and its cognitive correlates.

Table 4.1 Specific learning disorder (SLD): etiology, associated impairments, and cognitive correlates

SLD specifier	Subskill	Etiology	Associated impairments/cognitive correlates
With impairment in reading	*Word reading accuracy*	Several cortical and subcortical structures are frequently implicated, including the planum temporale, temporal lobes, corpus callosum, and cerebellum (e.g., Eckert et al., 2003). More recent work appears to identify dysfunction in a left hemispheric network that includes the occipitotemporal region, inferior frontal gyrus, and inferior parietal region of the brain (Silani et al., 2005; Shaywitz et al., 2000; Fletcher, Simos, Papanicolaou, & Denton, 2004; Richlan et al., 2009; Richlan, 2012). Numerous imaging studies have also found that dysfunctional responses in the left inferior frontal and temporoparietal cortices play a significant role with regard to phonological deficits (Skeide et al., 2015) Family and genetic factors have long been identified as crucial in dyslexia, with some researchers suggesting that a child with a parent with a reading disability is eight times more likely to be dyslexic compared to the general population (Pennington & Olson, 2005). Certainly, there is converging evidence from family and twin studies demonstrating the heritability and familiality of dyslexia (Grigorenko, 2001). Recently, genetic linkage studies have also identified several susceptibility genes for reading disabilities. These include sites on chromosomes 1, 2, 3, 4, 6, 11, 15, and 18, with one of the most commonly identified genetic loci being on chromosome 6 (Grigorenko, 2005; Paracchini et al., 2007; Scerri & Schulte-Korne, 2010; Scerri et al., 2011; Skeide et al., 2015) Shared environmental factors include language and literacy environment during childhood (Wadsworth et al., 2000) and quality of reading instruction	*Phonological awareness* – primary cognitive correlate; the metacognitive understanding that words have internal structures based on phonemes (Fletcher et al., 2007; Melby-Lervåg, Lyster, & Hulme, 2012; Willcutt et al., 2013). When this awareness is impaired, word recognition is delayed and fluency and comprehension skills are consequently affected *Rapid naming* – some researchers have found that phonological awareness and rapid letter naming both uniquely predict word recognition skills (Schatschneider, Fletcher, Francis, Carlson, & Foorman, 2004; Wagner, Torgesen, & Rashotte, 1994; Wagner, Torgesen, Rashotte, & Hecht, 1997). However, a meta-analysis of studies examining the relationship between rapid naming and dyslexia found little evidence to support a central and persistent deficit in naming speed in individuals with the disorder (Vukovic & Siegel, 2006). On the other hand, there are findings to suggest that phonological awareness and rapid naming, although correlated, are distinct variables and contribute uniquely to word recognition (Petrill, Deater-Deckard, Thompson, DeThorne, & Schatschneider, 2006) *Phonological memory* – working memory for verbal and sound-based information has also been found to be significantly related to word recognition, although it may not uniquely contribute when phonological processing is accounted for (Melvy-Lervag, Lyster, & Hulme, 2012; Schatschneider et al., 2004; Wagner et al., 1997; Willcutt et al., 2013)

(continued)

Table 4.1 (continued)

SLD specifier	Subskill	Etiology	Associated impairments/cognitive correlates
	Reading comprehension	Several brain regions are often implicated in reading comprehension. These include the anterior temporal lobe, inferior temporal gyrus, inferior frontal gyrus, inferior frontal sulcus, and middle and superior frontal and temporal regions (Ferstl et al., 2008; Gernsbacher & Kaschak, 2003). More recent research has revealed a relationship between listening and reading comprehension and activation along the left superior temporal sulcus, which has referred to by some as the "comprehension cortex" (Berl et al., 2010). However, broader pathways are also activated in reading comprehension, reflecting increased cognitive demand compared to listening comprehension. Genetic factors are said to account for 41 to 76 percent of the variance in comprehension (e.g., Betjemann et al., 2008; Harlaar, Dale, & Plomin, 2007; Petrill et al., 2007). While genetic factors that influence decoding and listening comprehension account for nearly 40 percent of the variance in reading comprehension, there is little evidence for an independent source of genetic influence on comprehension alone (Harlaar et al., 2010; Keenan et al., 2006). However, estimating the genetic influences on reading comprehension may be particularly sensitive to the type of assessment test used (Betjemann, Keenan, Olson, & DeFries, 2011)	*Oral language* – difficulties in reading comprehension are frequently associated with deficits oral language in general, including areas such as vocabulary, morphology, and syntax (Catts et al., 1999; Cutting & Scarborough, 2006; Share & Leikin, 2004; Torgesen, 2000; Willcutt et al., 2013) *Listening comprehension* – several studies have demonstrated that a unique portion of the variance in reading comprehension can be explained by listening comprehension (Cutting & Scarborough, 2006; Kendeou, van den Broek, White, & Lynch, 2009) *Working memory* – comprehension involves holding words and sentences in awareness, while integrating prior knowledge with incoming information (Carretti et al., 2009). Poor comprehenders may have particular difficulty updating/revising information already in working memory (Pelegrina et al., 2014) *Executive functioning* – several executive functions are involved in reading comprehension, including planning, organization, and self-monitoring (Cutting et al., 2009; Locascio, et al., 2010; Sesma et al., 2009). Weaknesses in these executive functions result in difficulties with higher-order comprehension skills such as inferencing and integrating prior knowledge, monitoring comprehension, and adapting to text structure or genre (Fletcher et al., 2007).

	Reading rate or fluency	Brain regions activated are similar to the network implicated in word reading, but additional activation is observed in areas involved in eye movement and attention (Jones, Ashby, & Branigan, 2013). Further, there is also evidence for increased activation in the left occipitotemporal region, in particular the occipitotemporal sulcus, which is important for rapid processing of letter patterns (Shaywitz et al., 2004; Dehaene & Cohen, 2011). Some studies have found increased activation in this region when normal reading automaticity is disrupted (Benjamin & Gaab, 2012) While limited, there is evidence of genetic influences specific to rapid naming and reading, suggesting that RAN may be etiologically distinct from phonological awareness (Byrne et al., 2005; Compton et al., 2001; Petrill et al., 2006). Genetic linkage studies have identified susceptibility genes for fluency, namely, chromosome 2 (Raskind et al., 2005)	*Rapid automatized naming (RAN)* – while the exact relationship between RAN and reading remains unclear, RAN is believed to be one of the best predictors of reading fluency (Georgiou et al., 2008, Tan et al., 2005). The automaticity required to complete RAN tasks is related to the ability to synthesize and automatize letter sequences/words when reading (Norton & Wolf, 2012). There are also a variety of cognitive processes implicated in rapid naming. These include attention, executive functions (i.e., response inhibition, set shifting), lexical retrieval, and processing speed (Moll, Gobel, & Snowling, 2015) *Orthographic processing* – processing of orthographic information (i.e., the ability to process units of words based on visual long-term memory representations) is considered critical in automatic word recognition and consequently plays a crucial role in fluency (O'Brien et al., 2011). This ability is often impaired or underdeveloped in some reading-disabled individuals
With impairment in mathematics	*Number sense*	Researchers differentiate between the basic processing of numerical information and processes involved in math calculation and problem-solving, suggesting that these are both structurally and functionally distinct (Ansari, 2010). The intraparietal sulcus in both hemispheres is widely viewed as crucial in processing and representing numerical quantity, although there may be differences in activation as a function of age (Ansari & Dhital, 2006; Ansari, Garcia, Lucas, Hamon, & Dhital, 2005; Dehaene et al., 2004; Kaufmann et al., 2006; Kucian, von Aster, Loenneker, Dietrich, & Martin, 2008; Price & Ansari, 2013; Mussolin et al., 2010a)	*Number representation* – math disorders are associated with weaknesses in fundamental number representation and processing, which manifest in difficulties with quantifying sets without counting, using nonverbal processes to complete simple numerical operations, and estimating the relative magnitude of sets (Geary, 2010, 2013; Geary et al., 2012; Geary et al., 2008; Geary et al., 2009; Halberda et al., 2008; Rouder & Geary, 2014; Feigenson, Dehaene, & Spelke, 2004; Mazzocco, Feigenson, & Halberda, 2011) *Number comparison* – several studies have indicated that math difficulties are associated with deficient basic number-processing abilities, such as number comparison (Price & Ansari, 2013). These weaknesses are characterized by increased reaction times and error rates on tasks that involve comparing numbers, with particular difficulty when numbers are closer together (Mussolin, Mejias, & Noel, 2010b)

(continued)

Table 4.1 (continued)

SLD specifier	Subskill	Etiology	Associated impairments/cognitive correlates
	Memorization of arithmetic facts	A left hemisphere network that includes the precentral gyrus, inferior parietal cortex, and intraparietal sulcus is often implicated in math fact retrieval (Dehaene & Cohen, 1994; Dehaene & Cohen, 1997; Dehaene et al., 1999). Further, some researchers believe that rote math facts are retrieved from verbal memory, thereby requiring activation of the angular gyrus and other regions associated with linguistic processes (Dehaene, 1992; Dehaene & Cohen, 1995; Dehaene et al., 1999)	*Long-term retrieval* – weak or impaired long-term retrieval of facts and increased error rates in recall (Geary, 1993; Mazzocco, Devlin, & McKenney, 2008). Because fact-retrieval mechanisms fail to develop adequately, fluency is impaired and those with dyscalculia continue to utilize procedural strategies rather than memory-based strategies (Geary, Bow-Thomas, & Yao, 1992; Geary, Hamson, & Hoard, 2000; Jordan & Hanich 2003; Hanich et al., 2001; Landerl, Bevan, & Butterworth, 2004)
	Accurate or fluent calculation	Regions of the left frontoparietal cortex, including the intraparietal sulcus, angular gyrus, and supramarginal gyrus, have been consistently associated with math calculation (Ansari, 2008; De Smedt, Holloway, & Ansari, 2011; Dehaene, Molko, Cohen, & Wilson, 2004; Dehaene et al., 2004). However, there is evidence to suggest that math fluency, while related to other skills, may be genetically distinct and may reflect variance above and beyond untimed calculation abilities (Hart, Petrill, & Thompson, 2010; Petrill et al., 2012). The dorsolateral prefrontal cortex has also been found to show increased activation during calculation, implying that executive functioning and working memory may be playing a role in the process (Davis et al., 2009)	*Long-term retrieval* (see above) *Rapid naming* – the rate of access to information in long-term storage is believed to affect calculation fluency (D'Amico & Passolunghi, 2009). Some studies have found that math disorders are associated with deficits in rate of access of numerical information alone (e.g., D'Amico & Guarnera, 2005), while others have demonstrated that rate of access to both numerical and nonnumerical information is impaired (e.g., Temple & Sherwood, 2002) *Processing speed* – there is a body of evidence to support the contribution of processing speed in math calculation fluency; however, the relationship remains unclear, as processing speed is often highly related to working memory and general intelligence (Berg, 2008; Bull & Johnston, 1997; Geary, 2011; Mazzocco & Rasanen, 2013; Willcutt et al., 2013)

	Accurate math reasoning	As mentioned above, the intraparietal sulcus is often identified as a neural correlate of math disorders. However, it is likely that an entire network of brain regions is implicated, as the intraparietal sulcus plays a role in a variety of cognitive processes involved in math achievement (Szucs & Goswami, 2013). It has been suggested that the parietal network is involved in manipulating numerical quantities (Lemer et al., 2003). Further, some studies have found that individuals with dyscalculia have structural abnormalities in the parietal cortex (Rotzer et al., 2008; Rykhlevskaia et al., 2009) Prevalence of math disabilities is about ten times higher in those with family members who had math disabilities (Shalev et al., 2001). Twin studies suggest a moderate genetic influence, with some studies finding additive genetic influences shared between math calculation and problem-solving and several working memory components (Kovas et al., 2007; Lukowski et al., 2014) Environmental factors, including motivation, emotional functioning (e.g., math anxiety), and suboptimal or inadequate teaching, may also contribute to math difficulties (Szucs & Goswami, 2013; Vukovic et al., 2013). Further, math achievement in particular may be associated with cultural or gender-based attitudes that may be transmitted in the family environment (e.g., Chiu & Klassen, 2010; Gunderson et al., 2011)	*Working memory* – because mathematical reasoning relies on concurrently retaining multiple pieces of information while performing one or more procedures or mental operations, working memory is often implicated. Those with math difficulties tend to struggle with holding information in working memory, updating or revising the information, and tracking or monitoring the process, resulting in difficulties in sequencing, increased errors in counting, and other procedural errors (Geary, 2003; Lukowski et al., 2014; Pelegrina et al., 2014; Peng & Fuchs, 2014; Raghubar, Barnes, & Hecht, 2010; Swanson & Jerman, 2006; Willcutt et al., 2013) *Visual-spatial ability* – visual-spatial skills, such visual perception, spatial reasoning, and mental rotation, have been found to influence math performance (Gunderson et al., 2012). Weaknesses in these may present as difficulties with representing numbers and aligning numerals and problems in areas such as geometry or fractions (Geary, 2004; Swanson & Jerman, 2006) *Attention and executive functioning* – math difficulties often reflect weaknesses in executive functioning skills, such as set shifting and cognitive inhibition (D'Amico & Passolunghi, 2009; van der Sluis, de Jong, & van der Leij, 2004; Willcutt et al., 2013). Further, poor attentional control (i.e., difficulty ignoring irrelevant information and focusing on goal-relevant information) is often observed (Geary, 2013)

(continued)

Table 4.1 (continued)

SLD specifier	Subskill	Etiology	Associated impairments/cognitive correlates
With impairment in written expression	*Spelling accuracy*	Functional neuroimaging studies have provided substantial evidence for the role of the ventral-temporal inferior frontal gyrus and the posterior inferior frontal gyrus in spelling (Rapp et al., 2015; van Hoorn et al., 2013). Other areas that have been identified include the left ventral cortex, bilateral lingual gyrus, and bilateral fusiform gyrus (Planton et al., 2013; Purcell et al., 2014; Richards et al., 2005; Richards et al., 2006). However, many of these regions have also been associated with reading and are not distinct to spelling/writing disorders There is evidence that links spelling to a region of chromosome 15 (Schulte-Korne, 2001), although this locus has also been reported in dyslexia (Grigorenko, 2005)	*Phonological processing* – phonological awareness is a significant predictor of spelling achievement (Caravolas, Hulme, & Snowling, 2001; Cornwall, 1992; Holm, Farrier, & Dodd, 2008; Skeide et al., 2015; Yeong, Fletcher, & Bayliss, 2014). Weaknesses in this area may manifest as poor segmentation of words into phonemes, poor sequencing of sounds, and omission or addition of sounds (Berninger, 1999) *Orthographic processing/orthographic coding* – effective spelling involves storing and retrieving commonly occurring letter patterns in visual and motor memory; these skills are often impaired in poor spellers (Caravolas, Hulme, & Snowling, 2001; Ehri, 2014; Yeong, Fletcher, & Bayliss, 2014) *Motor skills* – poor spelling is often accompanied by underlying skill deficits in areas such as fine-motor control, motor planning, orthographic motor coordination, and visual-motor integration (Christensen, 2004; Daly, Kelley, & Krauss, 2003; Feder & Majnemer, 2007)
	Grammar and punctuation	With regard to English grammar, some researchers distinguish between the mental lexicon (i.e., memorized associations) and mental grammar (i.e., language rules and structure) and posit that each has distinct neural correlates (Pinker, 1994). There is some evidence to support this view, with data indicating that the mental lexicon involves left temporal and temporoparietal regions, while the mental grammar recruits a system that includes left frontal regions (Ullman et al., 2005)	*Long-term memory* – it has been suggested that some components of long-term storage, in particular procedural and declarative memory, may be involved in grammar; however, much of this research has focused on children with language impairments (Conti-Ramsden, Ullman, & Lum, 2015; Hedenius, et al., 2012)

	Clarity of written expression	Neural correlates of writing are less understood, but some studies have suggested that the cerebellum and parietal cortex, particularly the left superior parietal lobe, may be involved (Katanoda et al., 2001; Magrassi et al., 2010). In addition, the frontal lobes have also been implicated and are considered crucial in planning, brainstorming, organizing, and goal setting (Shah et al., 2013) While there is a significant genetic component involved in the development of writing skills, this etiology is often shared with a broad variety of reading and language skills (Olson et al., 2013)	*Working memory* – a substantial body of research has highlighted the role of working memory in written expression, as text generation requires the coordination of multiple processes, such as synthesizing multiple ideas, retrieving grammar rules from long-term storage, and ongoing self-monitoring (Berninger, 1999; Bourke et al., 2013; Hooper et al., 2002; McCutchen, 1996) *Attention and executive functioning* – a variety of executive functions, including attention, planning, and self-monitoring, have been implicated in written expression (Altemeier, Jones, Abbott, & Berninger, 2006; Graham, Gillespie, & McKeown, 2013; Graham & Harris, 2005; Harris & Graham, 2013; Hooper et al., 2002; Mason, Harris, & Graham, 2011; Reiter, Tucha, & Lange, 2005; Rosenblum et al., 2009; Troia & Graham, 2002) *Language* – level of knowledge of syntax, morphology, semantics, and vocabulary has a significant impact on text generation ability (Dockrell, Lindsay, & Connelly, 2009; Fey, Catts, Proctor-Williams, Tomblin, & Zhang, 2004; Olinghouse & Wilson, 2013). Language impairments are associated with higher rates of grammatical errors, less lexical diversity, and poorer overall content (Fey et al., 2004; Mackie & Dockrell, 2004)

There is evidence to suggest the role of heredity as a significant contributory factor in the etiology of learning disabilities. Such evidence includes the fact that learning disabilities appear to run in families. Family and twin studies demonstrate moderate to high familiality and heritability of learning disabilities, specifically impairments in reading and spelling (Scerri & Schulte-Korne, 2010; Schulte-Korne, 2001). For example, heritability estimate values greater than 0.6 have been found for manifestations of learning abilities and disabilities (see Cortiella & Horowitz, 2014). Moreover, several genetic linkage studies implicate chromosomes 6 and 15 in the etiology of reading disorders, with chromosome 6 also suspected in the etiology of ADHD (Schulte-Korne, 2001; Willcutt, et al., 2002; see Table 4.1 for more information on heritability).

In addition to the role of genetics, prenatal and perinatal factors are considered to be contributory factors in the etiology of learning disabilities. Prematurity, low birth weight, and in utero exposure to alcohol or cocaine are all associated with increased risk for learning problems. Loss of oxygen during birth may also result in altered brain development contributing to low academic achievement. Maternal metabolic conditions, including diabetes, hypertension, and obesity, also appear to contribute to the development of neurodevelopmental conditions such as autism and learning problems (Krakowiak et al., 2012).

In addition, research from the 1990s and early 2000s supported neurobiological factors in the etiology of learning disabilities. In numerous publications, Shaywitz and Shaywitz (e.g., 2004) describe over-activation of Broca's area with under-activation in the left parietotemporal and occipitotemporal regions of the brain in children with reading disabilities. The left inferior prefrontal cortex, left angular gyrus, and inferior parietal lobes have been implicated in the development of math-related learning disabilities (Chochon, Cohen, van de Moortele, & Dehaene, 1999; Kesler, Sheau, Koovakkattu, & Reiss, 2011). In addition, the intraparietal sulcus appears to be a crucial region in the development of number sense (Dehaene, Molko, Cohen, & Wilson, 2004).

Furthermore, environmental characteristics are also considered to be etiological factors in the development of learning disabilities. Low socioeconomic status, low maternal education, male gender, and nonwhite race were all found to be related to increased risk for learning disabilities in a kindergarten sample (Resnick et al., 1999). Malnutrition may also be another significant environment factor contributing to learning problems (Groce et al., 2014).

Symptom Presentation

One essential feature of SLD, as set forth in Criterion A, is persistent difficulties learning vital academic skills that are first experienced during formal schooling. While skills like walking and talking typically develop naturally, academic skills require teaching and explicit learning. In the case of SLD, the normal pattern of acquiring academic skills is disrupted. Difficulties mastering basic skills such as reading, math, and writing can impact academic performance in other areas such as science and social studies. In addition, the learning difficulties persist for a period of 6 months or longer without evidence of "catching up" to same-grade peers despite provision of extra support. A review of educational history including school reports, work samples, curriculum-based measures, or clinical interview can establish the persistence of learning difficulties.

Another critical component of DSM-5 diagnosis of SLD is performance in an academic domain that is "well below average" for the individual's age, as set forth in Criterion B. Clinical indicators are below average scores in school or average performance with extraordinarily high levels of support, avoidance of activities requiring the impaired academic skill, as well as psychometric evidence from individually administered, psychometrically sound, norm-referenced, and culturally appropriate academic achievement measures. The authors of the DSM-5 description of SLD note that academic skills are distributed along a continuum, and, therefore, there is no natural cutoff score to

differentiate those who have an SLD from those who do not. Attempts to establish cut points are arbitrary, but the recommended 1.5 standard deviations below the mean are "needed for greatest diagnostic certainty." The authors provide a more "lenient threshold" of 1–2.5 (i.e., 85 to 63 standard score) standard deviations below the mean when learning difficulties are supported by other sources of evidence (APA, 2013, p. 69).

According to DSM-5, the third core feature detailed in Criterion C is that the learning difficulties become observable during the early school years in most individuals. However, the authors note that some individuals might not fully manifest symptoms of the disorder until learning demands surpass the individual's ability to compensate for "limited capacities." Thus, an adult manifesting low academic achievement in college, for example, may be diagnosed with an SLD for the first time.

Finally, Criterion D excludes learning difficulties attributable to other more general disorders such as intellectual disability (or intellectual developmental disorder, as it is termed in DSM-5). The DSM-5 considers "normal intellectual functioning" to be an IQ estimate above 70, give or take five points to account for measurement error, and such cognitive functioning is considered to differentiate an SLD from more general learning problems. In order to be considered to have an SLD, an individual must demonstrate "unexpected academic underachievement," which is cited as the defining characteristic of SLD. Adequate academic functioning may be able to be maintained with intense external supports, extraordinary effort, or compensatory strategies. Individuals who are intellectually gifted may also demonstrate symptoms warranting an SLD diagnosis. Criterion D also excludes general learning difficulties secondary to economic disadvantage, chronic absences, or lack of adequate education and to neurological, motor, vision, or hearing disorders. The learning difficulties are also "specific" in that they may be restricted to one academic skill or domain. The DSM-5 requires a comprehensive assessment involving professionals with expertise in SLDs and psychological/cognitive assessment. Diagnosis can only be made after formal schooling has begun, but can be made for an individual at any point during the life span provided evidence of difficulties during formal schooling is available. The clinical diagnosis must be made as a result of synthesizing medical, developmental, educational, and family history; the history of the academic problem; the impact of the problem on academic, occupational, or social functioning; previous and/or current school reports; work samples; curriculum-based assessments; and previous or current scores from individualized standardized tests of academic achievement (APA, 2013).

Associated Impairments and Comorbidities

Delays in language, attention, or motor skills often but not always precede the manifestation of SLD. In addition, individuals with SLD often demonstrate uneven profiles of abilities and perform poorly on psychological tests of cognitive processing. According to the DSM-5, "…it remains unclear whether these cognitive abnormalities are the cause, correlate, or consequence of the learning difficulties" (APA, 2013, p. 70). While cognitive deficits associated with reading (i.e., dyslexia) are well known, those deficits associated with mathematics disorders or written expression disorders are less well understood. The DSM-5 further notes that processing deficits observed in individuals with SLD are seen in individuals with other disorders as well, including ADHD and autism spectrum disorder (ASD).

In fact, the diagnosis of SLD is often comorbid with other types of neurodevelopmental disorders or other mental disorders. For example, children with ADHD are three times more likely than non-ADHD peers to have a learning disability, but estimates vary widely depending on type of disorder and criteria used for diagnosing learning disabilities (DuPaul, Gormley, & Laracy, 2013). While diagnosis of other disorders does not preclude the additional diagnosis of SLD, differential diagnosis under those conditions may be more difficult as other disabilities may account for learning problems. Clinical judgment is needed to

determine whether academic skill deficits set forth in Criterion A may be attributable to other disorders. For more information on impairments that are associated with SLD, see Table 4.1.

Course and Developmental Changes

As stated earlier, emergence of symptoms of SLD typically occurs within the developmental period during formal schooling, despite the fact that language or fine-motor delays may be evident earlier. The disorder is one that is lifelong, but symptom expression may vary depending on environmental demands on deficient skills, severity of the deficiency, comorbidity, and the supports and interventions provided to the individual. Typically, problems persist into adulthood (APA, 2013).

Age-related manifestations of symptoms of SLD vary. Preschool children may have difficulty speaking clearly, learning letters or numbers, or recognizing their name. Similarly, kindergarten children may have difficulty learning and/or writing the alphabet, writing their names, and so forth. Phonetic analysis may also be challenging for kindergarten students with SLD, and they may have trouble breaking words down into syllables. They may also have difficulty rhyming. As these children progress through the early elementary school years, they may have difficulty with sound-symbol correspondence, sounding out words, recognizing irregular words, or learning math facts. Children with SLD often perform academic tasks with significant effort or inefficiency (e.g., Geary, Hamson, & Hoard, 2000).

As children with SLD enter the late elementary and early middle school years, they may omit syllables in multisyllabic words and may have difficulty remembering information learned in class. In addition, they may demonstrate difficulty with comprehension, spelling, or writing assignments (Feifer, 2011; Mather & Wendling, 2011; Santangelo & Graham, 2014). Adolescents may have mastered basic reading skills such as decoding, but reading fluency may remain slow and effortful. They may demonstrate poor understanding of written text and may have difficulty with math problem-solving. For adolescents with poor reading skills, they may also demonstrate increasing difficulty in content areas such as science and social studies. Adolescents and adults may continue to demonstrate poor spelling and difficulty pronouncing multisyllabic words. Rereading in order to understand the text may often be necessary for older individuals with SLD. Adults may have difficulty with drawing inferences from written text or from numerical information. As a result, they may avoid tasks involving these skills; symptoms of anxiety may also accompany the expression of learning difficulties (APA, 2013).

Prognosis

The risk of SLD is increased by several environmental factors, including prematurity, low birth weight, and prenatal exposure to nicotine. More students with SLDs come from households living in poverty as compared to students from the general population. Students with SLDs are also more likely to be in foster care or homeless. Genetic and physiological factors also increase the risk of SLD. For example, the risk of SLD in reading and mathematics is 4–8 and 5–10 times higher, respectively, in first-degree relatives with these disorders compared to individuals without these disorders. Children whose parents have dyslexia or a history of reading difficulties are more likely to experience learning difficulties, literacy problems, or SLD (Cortiella & Horowitz, 2014).

Several factors in the preschool years are predictive of later learning difficulties or SLD. For example, inattention is predictive of later difficulties in reading and math, including failure to respond to effective academic interventions (e.g., Bental & Tirosh, 2007). Also, speech or language impairments, or deficits in specific cognitive processes, such as phonological processing, working memory capacity, and naming facility in the preschool years, predict later SLDs (e.g., Scarborough, 2005). Furthermore, comorbidity with other disorders, such as ADHD, is predictive of worse mental health as compared to SLD without comorbidity (e.g., Smith & Adams, 2006).

Outcomes

Outcomes for individuals with SLDs depend on many variables, such as type of treatment, severity of the disorder, intensity of intervention, sustainability of treatment, nature and extent of comorbidities, effort and motivation of the individual, and availability of resources (e.g., parent involvement, home-school collaboration, accommodations). Table 4.2 provides a summary of outcomes for students with SLDs. It is important to note that poor outcomes may be circumvented by evidence-based interventions that are systematic, intensive, and individualized.

Intervention

The primary purpose of intervention for individuals with SLDs is *remediation* of skill deficits. Many interventions, particularly for reading, have been subjected to a rigorous evaluation and were found to be effective, meaning that when implemented with fidelity, they lead to positive outcomes (for a review see Cooney, Huser, Small, & O'Connor, 2007; Feifer, 2011, 2014; Flanagan & Alfonso, 2011; Kilpatrick, 2014; Mascolo, Alfonso, & Flanagan, 2014). Many of these interventions can be found at *Work Works Clearinghouse* (www.ies.ed.gov/ncee/wwc).

Table 4.2 Summary of academic performance and school outcomes for students with specific learning disorders

Twelve to twenty-six percent of secondary students with learning disabilities received average to above-average scores on math and reading assessments, compared to 50% of students in the general population
Seven to twenty-three percent of secondary students with learning disabilities receive well below average scores on academic performance, compared to 2% of students in the general population
Students with learning disabilities earn lower grades and experience higher rates of course failure in high school than students without learning disabilities
About one-third of students with learning disabilities have been retained at least once
One in every two students with a learning disability faced a school disciplinary action such as suspension or expulsion in 2011
Sixty-eight percent of students with learning disabilities leave high school with a regular diploma, while 19% drop out and 12% receive a certificate of completion
Sixty-nine percent of students with learning disabilities have failed one or more graded courses in secondary school, compared to 47% of students in the general population
Black and Hispanic students with disabilities experience much higher rates of school disciplinary actions, higher rates of dropout, and lower rates of graduation

Source: Cortiella & Horowitz (2014)

Not surprisingly then, evidence-based interventions are often the ones that are used first in either general or specialized instructional settings as compared to those interventions and techniques without such support. In general, it is incumbent upon practitioners to use evidence-based interventions with students who struggle academically. It is also prudent to use comprehensive interventions that can meet students' multiple manifest academic difficulties (e.g., remedial reading programs that contain the five essential components of reading; Feifer, 2011). However, it is clear from the literature that despite their overt relevancy, not all comprehensive, evidence-based interventions address the academic needs of every student effectively (e.g., Della Toffalo, 2010; Hale, Wycoff, & Fiorello, 2011).

In a tiered service delivery model, interventions are selected based on universal screening data. For example, students who are at risk for reading difficulties may receive *Wilson* if their reading difficulties are related primarily to decoding difficulties or *Read 180* if their reading difficulties are related primarily to comprehension difficulties (e.g., Feifer, 2011). When a student does not respond as expected to evidence-based interventions, a comprehensive evaluation is often recommended to gain a better understanding of the nature of and basis for the student's learning difficulties. Based on our knowledge of and expertise in evaluation of specific learning disabilities, we are of the opinion that it is only through a comprehensive and focused evaluation of cognitive abilities and processes and specific academic skills that the intervention process moves from *selecting an intervention program* to *tailoring an intervention* to meet the needs of students who do not respond as expected to evidence-based instruction (Mascolo, Flanagan, & Alfonso, 2014).

Selecting Interventions Versus Tailoring Interventions

Selecting interventions involves identifying evidence-based interventions that are most often used in standard service delivery models to address manifest academic difficulties that are revealed via progress monitoring (e.g., a particular reading program is selected by a district as a Tier II intervention for students with reading fluency difficulties). On the other hand, a primary focus of *tailoring* interventions involves understanding the student's pattern of cognitive and academic strengths and weaknesses and how this pattern interacts with the instructional materials used by the student as well as classroom instructional factors, environmental factors, and other individual/situational factors that may facilitate or inhibit learning. The goals for tailoring intervention, therefore, are (a) to use information about a variety of factors that are intrinsic and extrinsic to the student to design specific interventions and (b) to ensure that a student has appropriate access to the curriculum by minimizing or bypassing the adverse effects that cognitive and other weaknesses have on the student's learning. Tailoring interventions may include modification (e.g., instructional, curricular), accommodation, remediation, and compensation. Definitions and examples of these interventions are provided in Table 4.3. For additional information about these interventions and the evidence in support of them, see Mascolo, Alfonso, and Flanagan (2014).

Vagaries in the DSM-5 Criteria for Specific Learning Disorder

Historically, clinicians over-relied on IQ scores when making decisions about specific learning disorders and disabilities. A large body of

Table 4.3 Methods of tailoring intervention for students with learning disorders

Tailoring method	Brief description	Examples
Modification	Changes content of material to be taught or measured; typically involves changing or reducing learning or measurement expectations; may change the depth, breadth, and complexity of learning and measurement goals	Reducing the amount of material that a student is required to learn Simplifying material to be learned Requiring only literal (as opposed to critical/inferential) questions from an end-of-chapter comprehension check Simplifying test instructions and content
Accommodation	Changes conditions under which learning occurs or is measured, but does not change or reduce learning or assessment expectations. Accommodations may include timing, flexible scheduling, presentation, setting, and response accommodations	Extending time on exams Assigning a project in advance or allowing more time to complete a project Aligning math problems vertically, as opposed to horizontally Providing a separate room to work Having a student dictate responses to a scribe
Remediation	Techniques or programs used to ameliorate cognitive and academic deficits. Academic interventions typically focus on developing a skill, increasing automaticity of skills, or improving the application of skills. Cognitive interventions typically focus on improving cognitive processes such as working memory capacity and phonological processing. There are many techniques, published programs, and software designed for the purpose of remediation	Evidence-based programs listed at What Works Clearing House: http://ies.ed.gov/ncee/wwc Reading programs appearing on the Florida Center for Reading Research website: www.fcrr.ord Techniques and materials from the Reading Rockets website: www.readingrockets.org CogMed (Pearson) Spotlight on Listening Comprehension (LinguiSystems, 2006)
Compensation	Procedures, techniques, and strategies that are intended to bypass or minimize the impact of a cognitive or academic deficit	Teaching the use of mnemonic devices Organizational aids or techniques Teaching a student to outline or use graphic organizers

research has shown that the IQ-achievement discrepancy method, when used as a primary or sole criterion for diagnosis of SLD, is invalid (see Stanovich, 2005, for review). Therefore, the developers of the DSM-5 criteria for SLD should be commended for dropping this criterion. However, it seems clear from existing research that assessment of specific cognitive abilities and processes is an invaluable component to evaluation of suspected learning disorders and disabilities (see Hale et al., 2010). As such, DSM-5 criteria for SLD diagnosis are limited and when applied in isolation may obscure important information necessary to understand the nature of learning problems and lead to overdiagnosis. Following are a few of the most salient vagaries in DSM-5 criteria for SLD.

First, the DSM-5 definition of SLD relies on below average achievement without reference to underlying cognitive processing weaknesses that are known to interfere with basic academic skill acquisition and development. Below average achievement is caused by a variety of factors (e.g., attention problems, depression, poor motivation, behavior problems, low IQ), and the failure to include specific cognitive processes as part of the diagnostic criteria makes it difficult to determine the basis for academic underachievement. Essentially, the current definition reflects a "low achievement" model for defining and diagnosing SLD with an added caveat regarding poor response to intervention. Data show that states that rely on low achievement and poor response to intervention only (e.g., Iowa), like the DSM-5 criteria, have the highest percentage of students in special education under the category of specific learning disability. For example, while the national average of students with specific learning disabilities in special education is 41.5%, over 60% of the total special education enrollments in Iowa are students with specific learning disabilities (IDEAdata.org; cited in Cortiella & Horowitz, 2014).

Second, there are a number of terms and phrases in the DSM-5 criteria and diagnostic features that are not defined sufficiently. For example, DSM-5 criteria allow for diagnosis of SLD with average achievement scores if those scores are only attainable by "extraordinary effort." Because extraordinary effort is not defined objectively, its meaning is left up to the varied, unstandardized judgments of clinicians based largely on subjective data. Also, it is not clear what is meant by "exceed the individual's limited capacities," despite the examples that were given, including reading and writing lengthy complex reports and excessively heavy academic loads (see Criterion C). One problem with this criterion is that individuals with *average* cognitive and academic abilities often have difficulties with these tasks and often fail to meet the demands of an excessively heavy academic load, for example. This fact, coupled with the fact that the DSM-5 criteria allow for diagnosis of SLD in adulthood based on self-report or report by others, could potentially allow for just about anyone to be diagnosed with SLD, particularly older individuals who struggle in postsecondary educational programs.

Third, and perhaps most alarming, is the definition of "normal levels of intellectual functioning." It is important to know an individual's level of intellectual functioning because SLD is "specific" and, therefore, not attributable to intellectual disability (or intellectual developmental disorder) or global developmental delay, for example. According to the DSM-5 criteria for SLD, normal intellectual functioning is generally estimated by an IQ score of greater than about 70 ± 5 points (or standard scores of 65–75 on standardized tests having a mean of 100 and standard deviation of 15). This score range, which corresponds to the 1st to 5th percentiles, can in no way be conceived of as reflecting "normal" intellectual functioning. In fact, this score range is associated with *cognitive impairment* and *intellectual disability*. This score range is also identical to the range used to define intellectual developmental disorder in the DSM-5 (APA, 2013, p. 37).

Interestingly, the DSM-5 SLD criterion for academic skills that are "substantially and quantifiably below those expected for the individual's chronological age is "low achievement scores on one or more standardized tests or subtests within an academic domain (i.e., at least 1.5 standard deviations below the population mean for age, which translates to a standard score of 78 or less, which is below the 7th percentile)." Moreover,

standard scores in this range are "needed for greatest diagnostic certainty" (APA, 2013, p. 69). Using the properties of the normal probability curve, this criterion is certainly in line with what can be considered substantially below expected levels of performance. Therefore, it is neither logical nor accurate to interpret scores that are lower than 78 (i.e., scores of 70 + 5) as representing "normal" (intellectual) functioning.

Because the same standard score range is used to define normal intellectual functioning for SLD and impaired intellectual functioning for intellectual developmental disorder, the defining characteristic of SLD – *unexpected underachievement* – is not evident in the DSM-5 criteria, despite mention of this hallmark characteristic in the "diagnostic features" section (APA, 2013, p. 69). For example, individuals with IQs in the 1970s and commensurate academic achievement scores meet DSM-5 criteria for SLD. However, this type of profile does not reflect unexpected underachievement, but rather *expected underachievement*, which historically has never been a defining characteristic of SLD. Expected underachievement is more consistent with general learning difficulties that are perhaps due to unknown conditions, such as fetal alcohol spectrum disorder (e.g., Kodituwakku, 2007). Because the DSM-5 criterion for normal intellectual functioning is inaccurate, the definition of SLD is overly sensitive but not sufficiently specific, which will undoubtedly lead to a substantial increase in the number of individuals diagnosed with SLD.

It is beyond the scope of this chapter to discuss all of the potential issues related to diagnosis of SLD brought on by the revised SLD criteria included in the DSM-5. It is our contention that failure to include criteria related to cognitive processes obscures the very nature of SLD. There is an abundance of evidence in the cognitive and neuropsychological literature demonstrating how cognition affects achievement (see Fiorello, Hale, & Wycoff, 2012; Fletcher-Janzen & Reynolds, 2008). Most academic learning tasks require general cognitive processes such as attention, language, and memory, and specific processes contribute uniquely to specific academic skills (see Geary, 2011; Shaywitz et al., 2004). Even the authors of the DSM-5 definition of SLD found in their review of predictive validators "quite strong support for the inclusion of cognitive processing deficits in the diagnostic criteria for SLD," especially in reading (Tannock, 2013, p. 18). Academic tasks require numerous cognitive processes. Administering specific narrowly defined cognitive tasks helps to systematically isolate the underlying learning problems and differentiate SLD from other conditions that affect learning and achievement (see Decker, Hale, & Flanagan, 2013, for a discussion). The purpose of administering cognitive tests is not necessarily to obtain an IQ score, but to assist in understanding learning problems (Flanagan et al., 2013) and to develop effective interventions (Fuchs et al., 2011).

In short, we believe that approaches to SLD diagnosis that do not consider cognitive processing strengths and weaknesses are not supported by research (see Reynolds & Shaywitz, 2009). We support alternative research-based approaches to SLD diagnosis "because they emanate from the marriage of a collective body of knowledge that has been acquired through research in the fields of neuroscience, pedagogy, assessment, and intervention" (Della Tafallo, 2010, pp. 180–181). These approaches de-emphasize IQ in favor of theory-based flexible batteries that include measures of cognitive abilities and processes that are predictive of specific academic skills and that yield information relevant for instructional planning (see Flanagan & Alfonso, 2011, for a review).

Conclusion

This chapter provided a brief history of the definition of specific learning disability with emphasis on the criteria included in the five editions of the DSM. The DSM-5 criteria were reviewed in detail. Also reviewed was the etiology of SLD as well as associated impairments and comorbidities, course and developmental changes, prognosis, outcomes, and treatment. This chapter also highlighted some salient unexpected changes in

the DSM-5 criteria, changes that appear to obscure the true nature of SLD as a disorder in one or more basic psychological processes.

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Part III

Internalizing Disorders: Schizophrenia Spectrum

Childhood-Onset Schizophrenia Spectrum Disorders

5

James Knorr

Schizophrenia and other schizophrenia spectrum disorders are chronic and debilitating psychiatric disorders that affect approximately 1% of the population (van Os & Kapur, 2009). Individuals with these disorders often experience significant impairments in daily functioning (Regier et al., 1993). The peak age of onset ranges from 10 to 25 years in males and 25 to 35 in women (Carpenter & Buchanan, 1994).

Childhood-onset schizophrenia (COS) is generally regarded as a schizophrenia disorder diagnosed before the age of 13. It is considered a rare and more severe form of schizophrenia compared to adult-onset schizophrenia (AOS). The estimated rate of occurrence is 1 in 10,000 children (Gonthier & Lyon, 2004). Of all schizophrenic disorders, only 0.1–1% manifest prior to the age of 10, increasing to 4% by the age of 15 (Dulmus & Smyth, 2000). COS is more prevalent in males, but this ratio tends to find balance as children age into adolescence. This is possibly due to the development of secondary sex characteristics in females associated with the onset of psychosis (Jacobsen & Rapoport, 1998; McClellan and Stock, American Academy of Child and Adolescent Psychiatry [AACAP], 2013).

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Behavioral and cognitive symptoms associated with COS overlap features of autism spectrum disorders (ASD), affective and disruptive behavior disorders, and speech and language disorders. Because of the debilitating nature of COS, it is crucial for an accurate diagnosis and appropriate treatments to be implemented. This chapter will provide an overview of the diagnostic history, symptomology, associated impairments, developmental course, prognosis, and treatments associated with this disorder.

Diagnostic History

In the *Diagnostic and Statistical Manual of Mental Disorders – 2nd Edition (DSM-II)*, COS was included under the category of schizophrenia, but there was not a distinction between autistic behaviors and behaviors currently associated with schizophrenia (American Psychiatric Association [APA], 1968). In the DSM-III, authors began to make a distinction between the two disorders, categorizing autistic behaviors as a pervasive developmental disorder (PDD) (APA, 1980). The DSM-IV-TR further delineated COS from PDDs in that pervasive developmental disorders are "characteristically recognized during infancy or early childhood (usually before age 3 years), whereas such early onset is rare" in schizophrenia (APA, 2000, p. 311). Furthermore, they asserted that PDDs often lack prominent

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delusions and hallucinations, and are characterized by more stereotypies and abnormalities in language.

In the current revision of the DSM, symptoms of COS are similar to those in AOS but may be more difficult to diagnose. "In children, delusions and hallucinations may be less elaborate than in adults and visual hallucinations are more common and should be distinguished from normal fantasy play" (APA, 2013, p. 102). Authors of the DSM-5 also recognize that disorganized speech and behaviors occur in many other childhood disorders and these more prominent disorders should be given more consideration.

DSM-5

In the DSM-5, COS is not regarded as a specific diagnosis. Rather it is included under the diagnostic category of schizophrenia (APA, 2013). The diagnostic criteria includes children who demonstrate the following behaviors across a significant portion of time:

1. Delusions
2. Hallucinations
3. Disorganized speech
4. Grossly disorganized or catatonic behavior
5. Negative symptoms

One of these behaviors must be delusions, hallucinations, or disorganized speech. Continuous signs of these behaviors must persist for at least 6 months with at least 1 month of significant symptoms. Periods of prodromal or residual symptoms may also be apparent. Like many other diagnoses in the DSM-5, one or more significant areas of the child's life must be affected. Specifically for COS, "when the onset is in childhood or adolescence, there is a failure to achieve expected level of interpersonal, academic or occupational functioning" (p. 99). Moreover, because of the rare and debilitating nature of COS, other disorders such as ASD and attention-deficit/hyperactivity disorder (ADHD) must be ruled out.

Symptom Presentation

The same diagnostic criteria are used for COS and AOS, but several distinctions should be made. COS is a rare form of schizophrenia and follows a different developmental course. Characteristic symptoms of schizophrenia (hallucinations, delusions, disorganized speech, etc.) should be distinguished from typical developmental behaviors or more common diagnoses. Moreover, impairment in functioning should be considered relative to a child's expected level of interpersonal, academic, or occupational functioning.

Positive symptoms of schizophrenia include, but are not limited to, hallucinations, delusions, thought distortions, and/or disorganized speech or behaviors. Hallucinations are an apparent perception of something that is not readily present. These may manifest auditorily or visually. Hallucinations may be striking, but must be considered within the developmental context of the child. Garralda (1984) reported that most children who report hallucinations do not meet the criteria for schizophrenia, nor have a psychotic illness. Imagination and fantasy should also be distinguished from auditory and visual hallucinations (Caplan, 1994). For example, children often develop an imaginary friend or believe that someone is talking to them when alone. Developmentally, an imaginary friend can serve as someone who has similar interests to the child while hearing voices may be the child's way of describing their covert self-talk.

Delusions can be characterized as idiosyncratic beliefs or impressions that are firmly maintained, despite being either contradicted, generally believed, or taken as reality in typical situations. These should also be considered within the developmental context of the child. Delusions including superstitious thoughts or developmentally appropriate beliefs should be ruled out through behavioral observations and communication with the child. An understanding of why such thoughts and beliefs are important in their life will be critical. For example, a developmentally appropriate delusion in a child may be that if they carry their favorite object or wear the same clothes daily, this will help their ill sibling recover.

Disorganized speech or behaviors may also be apparent. As with other symptoms, these must also be distinguished from typical childhood behaviors or more common childhood diagnoses. Language disorders, ADHD, ASD, obsessive-compulsive disorder (OCD), and other more common developmental disorders should be considered in addition to COS when formulating a diagnostic impression.

Negative symptoms may include a flat affect, lack of energy, catatonia, avolition, and/or depressed thoughts, among others. Negative symptoms are not required for a diagnosis of COS, but if present, they are often severe and cause impairment across several areas of functioning. These symptoms must also be distinguished from more commonly diagnosed disorders such as bipolar disorder, major depressive disorder, and other psychiatric disorders (American Academy of Child and Adolescent Psychiatry [AACAP], 1998; Werry, McClellan, & Chard, 1991).

Etiology

Genetics contribute to the increased risk of developing schizophrenia. In family studies, there has been shown to be a threefold increase in relative risk for developing schizophrenia in parents of schizophrenia probands (Kendler et al., 1993). Similarly, morbid risk for parents of individuals with AOS probands is 6% compared to 9% for siblings (Tsuang, Stone, & Faraone, 2000). In an early twin study, researchers found a concordance rate of 88.2% between monozygotic twins for a diagnosis of COS compared to 22.3% for dizygotic twins (Kallmann & Roth, 1956). This resulted in an estimated heritability of 84.5%.

In addition to schizophrenia, other psychiatric disorders have also been found to have an elevated relative risk for developing COS. Asarnow et al. (2001) found a relative risk of 10.5% and 5.6% for COS in children that have a parent diagnosed with a schizotypal personality disorder or avoidant personality disorder, respectively. Nicolson et al. (2003) found a relative risk of 15.2% for developing COS when having a parent with a paranoid personality disorder. Across these studies, these authors also found a relative risk of 16.9% and 15.9%, respectively, for developing COS with a parent diagnosed with a schizophrenia spectrum disorder.

Genetic deletions and duplications have been found common in individuals with schizophrenia spectrum disorders. While these seem to be rare genomic occasions and structural mutations, some have been implicated through their symptomology associated with a schizophrenia spectrum disorder. Most notably, 22q11.2 deletion syndrome (velocardiofacial syndrome) is associated with substantial rates of behavioral, cognitive, and psychiatric problems, including psychosis (Sporn et al., 2004). Other structural mutations at 1q21.1 and 15q13.3 have also been noted to contribute to symptoms associated with schizophrenia spectrum disorders. Although noteworthy, combined with the 22q11.2 mutation, these genomic "hotspots" may only be responsible for 0.5% to 1.0% of all cases of schizophrenia specturm disorders (McClellan et al., 2013, p. 979).

Neuroimaging studies have repeatedly found differences between individuals with COS and AOS when compared to the general population. Sprooten et al. (2015) and Greenstein et al. (2006) found cortical thinning in AOS and COS participants, respectively. When individuals with COS were compared to their nonpsychotic siblings, Mattai et al. (2011) found similar significant cortical thinning in the prefrontal, left temporal, right inferior temporal, and parietal areas; however, in their nonpsychotic full siblings, this thinning tended to normalize around the age of 17.

Deficits in the prefrontal and temporal cortex have been widely researched as possible contributors to symptoms associated with COS and AOS. Deficits may be a result of a pattern of thinning starting from the parietal lobe that continually spreads to the frontal and temporal areas (Yildiz, Borgwardt, & Berger, 2011). While individuals with AOS tend to have typical parietal lobes in neuroimaging studies, individuals with COS may have a pattern described as an "exaggerated form of neurodevelopmental 'pruning,' and may demonstrate

the neural correlate of one of the few distinguishing characteristics between COS and AOS. It seems as though COS patients undergo an early parietal deficit that is not so much abnormal as it is poorly timed; the normal maturational curve of parietal development is similar, yet delayed, compared to the COS population" (Ordóñez, Sastry, & Gogtay, 2015, p. 443). Prominent working memory deficits in individuals with schizophrenia have also been associated with prefrontal lobe deficits (Lett, Voineskos, Kennedy, Levine, & Daskalakis, 2014).

Temporal lobe volume abnormalities are consistently associated with temporal cortical gray matter decreases, associated with both emotional and auditory processing (Gogtay et al., 2004; Rapoport, Giedd, & Gogtay, 2012). Such deficits could possibly affect social functioning and be related to symptoms of social withdrawal and auditory hallucinations (Bunk, Eggers, & Klapal, 1999; Hugdahl, Loberg, & Nygard, 2009).

Neuroimaging studies have consistently shown larger ventricular sizes in individuals with COS and AOS when compared to the general population (Jacobsen et al., 1997; Mehler & Warnke, 2002). Such increases may be a distinctive feature of COS and AOS. Ventriclar size may contribute to decreased gray matter; yet strong physiological and psychological correlates of these two features have yet to be distinguished in the research literature (Olabi et al., 2011).

Subcortical areas have been researched but are still in need of further clarification as to their contributions to symptoms associated with COS. The right anterior insula is believed to contribute to processing visual and auditory emotional information, as well as to distinguish one's self from outside stimuli (Wylie & Tregellas, 2010). One study found slight decreases in left, right, and bilateral insula volumes in individuals with COS relative to controls. These may contribute to emotional processing deficits through visual and auditory modalities, as well as hallucinations and deficits in interoception (Moran et al., 2013).

The hippocampus and amygdala have been robustly studied in individuals with AOS, but research has been inconsistent or unable to be produced in individuals with COS (Kraguljac, White, Reid, & Lahti, 2013; Nugent et al., 2007). Giedd et al. (1999) found decreases in hippocampal volume in individuals with COS as they age into adulthood. Subsequently, abnormal amygdala activation and a decreased hippocampal volume have been noted in individuals with AOS (Pankow et al., 2013; van Erp et al., 2013). Due to the significant role the amygdala plays in the emotional and social processing of memory, further research using more precise imaging techniques will be helpful to provide more consistent findings.

Other subcortical areas associated with schizophrenia include the cerebellum and basal ganglia. Abnormal size, volume, and functioning in these areas have been found in individuals with AOS, but research with children has produced either inconsistent or insufficient results (Ordóñez et al., 2015). Due to the motor and sensory anomalies associated with COS and AOS, as well as the use of dopamine as a treatment modality, future research will be important for diagnostics and treatment planning.

Environment factors may contribute to, and affect, disease risk and progression. Maternal malnutrition, maternal age, prenatal infections, obstetric complications, and drug use are noteworthy for developing a schizophrenia spectrum disorder (McGrath & Susser, 2009). Research as to the effects of these, and other environmental factors, is limited at this time, and should be further explored.

Neuropsychological Implications

Kumra et al. (2000) found pediatric patients with either COS or a psychotic disorder – not otherwise specified (PD-NOS) were consistently one or two standard deviations below normative data on a broad array of cognitive abilities. Fifty-one participants, who were non-responsive to neuroleptic medication, were assessed with multiple measures of cognitive abilities and educational knowledge. Full scale IQ scores for COS and PD-NOS groups were below average, while performance on spelling and reading decoding assessments was average.

Biswas, Malhotra, Malhotra, and Gupta (2006) studied a sample of 15 participants with COS. Deficits were found in verbal intelligence, language knowledge, visual-spatial abilities, and perceptuomotor skills. Participants also demonstrated difficulty on abstraction and memory tasks. Similar to Kumra et al. (2000), participants demonstrated below average full scale IQ scores.

Biswas et al. (2006) also compared the neuropsychological profile of participants with COS to participants with adolescent-onset schizophrenia (AdOS) and participants with AOS. Similar neuropsychological profiles were found between COS and AdOS groups, but deficits were significantly greater in the COS group. Moreover, significantly improved performance was found on perceptuomotor functioning, most areas of memory, and full scale IQ scores (nearly 20 points greater and within the average range) in the AOS group compared to the other groups.

Neurological deficits tend to be more apparent and debilitating in individuals with COS compared to individuals with AOS. Because research is limited at this time, in-depth analyses of neuropsychological deficits should be examined further. How such deficits relate to functioning should also be explored.

Developmental Course

COS is considered a severe form of schizophrenia due to the age of onset of symptoms and its developmental course. Children often experience impairments in multiple life areas due to symptom severity. Positive outcomes in COS are similar to the protective factors associated with AOS. Average to above-average premorbid functioning, higher cognitive abilities, and predominately positive symptoms in females (hallucinations, delusions, etc.) are some protective factors. Risk factors include a delayed onset of symptoms (resulting in delay of diagnosis and treatment), onset prior to the age of 12, premorbid dysfunction, severe positive and negative symptoms, lower cognitive abilities, and poor interpersonal skills (McClellan, McCurry, Snell, & Dubose, 1999).

Protective and risk factors are correlated, and may serve as moderator variables to symptoms related to COS. For example, possessing strong cognitive abilities may result in better interpersonal skills, better premorbid functioning, and possessing more positive symptoms (Gonthier & Lyon, 2004). Conversely, children with lower intellectual abilities tend to demonstrate poorer premorbid functioning and experience more negative symptoms. Moreover, positive symptoms tend to alleviate over time while negative symptoms may persist, resulting in a longer duration of impairment.

Gonthier and Lyon (2004) described four phases that children with COS experience related to their symptomology. These phases tend to cycle. While children may not experience symptoms related to all phases, deterioration of functioning tends to increase with each additional cycle.

The first phase is the prodromal phase. This is characterized by a less severe functional deterioration before the onset of psychotic symptoms. Interpersonal skills tend to be negatively affected and may include social withdrawal and isolation. Internalizing symptoms such as depression and anxiety, conduct problems such as substance abuse, and aggressive behaviors may be present (McClellan et al., 2013). Symptoms may also include bizarre preoccupations, lack of self-care skills, sleeping difficulties, changes in appetite, or somatic symptoms. Academic withdrawal may also result from symptoms.

The acute phase is characterized by an onset of positive symptoms. These may result in a significant decrease in cognitive and social functioning. The severity of symptoms in this phase often lead to initial diagnoses and treatment. If support is quick, the presentation of positive symptoms may only last several months, depending in part on the child's response to treatment. If symptoms are more severe or resistant to treatment, hospitalization or residential treatment may be required.

The recuperative or recovery phase is the third phase. This phase is associated with an alleviation of some positive symptoms and a predominance of negative symptoms. Negative symptoms

are often severe, resulting in significant impairment. Children may not require an inpatient setting and may return to school, but often struggle with poor attention and internalizing symptoms. They may demonstrate a flat affect and bizarre behavior. Dysphoria may result in withdrawal from social situations and apathy.

The residual phase is characterized by an absence of positive symptoms. Negative symptoms may persist but are often less severe than in the recovery phase. Children often demonstrate an increase in functioning but still experience impairment relative to their premorbid functioning.

The four phases of COS tend to cycle. Children may experience symptoms related to the residual phase but then relapse back into a prodromal or an acute phase. With pharmacological treatment, relapses tend to prolong between cycles and positive symptoms may diminish over time (Leucht et al., 2012). Negative symptoms tend to remain stable or increase over time without psychotherapeutic treatments.

Prognosis/Outcome

Several longitudinal studies have examined the prognosis and outcomes of children with COS. These studies followed children diagnosed with COS into their early and mid-twenties to determine whether they still met the diagnostic criteria for schizophrenia and experienced functional impairments.

Due to persistent symptoms, children diagnosed with COS tend to meet the diagnostic criteria years after their initial diagnosis. In an early follow-up study of 18 participants diagnosed with schizophrenia or a schizoaffective disorder in childhood, Asarnow, Thompson, and Goldstein (1994) found that despite extensive pharmacological and psychosocial treatment, the majority of participants continued to demonstrate symptoms as they aged. Of their sample, 67% continued to meet the criteria for schizophrenia or a schizoaffective disorder, albeit 56% showed substantial improvement in their functioning at 2- to 7-year follow-up.

Hollis (2000) attempted to follow 110 subjects between 1973 and 1991 who experienced their first episode of psychosis in childhood or adolescence. Of the initial sample, 84.5% were successfully followed for an average of 11.5 years. There was a positive predictive value (PPV) of 80% for diagnostic stability of schizophrenia. The researchers also found a PPV of 83% for affective psychoses. Comparatively, patients who initially received diagnoses of a schizoaffective disorder or atypical psychoses had much lower PPVs. Overall, compared to the other psychoses studied, COS and AdOS were associated with significantly worse outcomes, characterized by chronic illness, impairment in social relationships, and dependent living.

Lay, Blanz, Hartmann, and Schmidt (2000) attempted to follow 96 inpatient subjects who met the ICD-9 diagnostic criteria for schizophrenia. In this study, 68% of the original sample was successfully followed at a 10-year follow-up. Eighty-three percent had at least one further inpatient treatment episode, while 74% were receiving psychiatric treatment at follow-up. Of the participants, 57% reported at least moderate impairment in vocational functioning, 66% reported serious social impairment, and 75% reported being financially dependent on parents or public assistance.

Between these two studies, the researchers found several areas of functioning that are consistently, and negatively, impacted by symptoms of COS. Lower educational achievement, financial and emotional dependence, and persistent outpatient, residential, or inpatient treatment were commonly found (Hollis, 2000; Lay et al., 2000).

Social relationships and employment are often negatively affected by symptoms of COS. Analysis of two long-term follow-up studies found that of adults who initially received diagnoses of COS or AdOS, only 7% reported being involved in satisfying partnerships (Eggers, Bunk, & Roepke, 2002). Eighteen percent reported being engaged to a significant other, 7% were divorced, and 59% never married and lived alone. With employment, 34% reported being unable to work or were unemployed, 25% considered themselves to be homemakers, and only 41% had some type of employment.

Risk of suicide has also been associated with first episode psychoses and schizophrenia spectrum disorders. Examining 149 participants with first episode psychoses, nearly 46% of participants were suicidal at the time of the study (Barrett et al., 2010). Moreover, at least 5% of individuals with COS die by suicide or accidental death related to psychotic thinking (Eggers, 1978; Werry et al., 1991).

Assessment

A comprehensive neuropsychological evaluation is paramount in diagnosing COS due to the overlap of symptoms with more prominent neurodevelopmental, mood, behavioral, and language disorders. Distinguishing symptoms of COS from more common mental disorders or typical childhood development should be a predominant goal for clinicians.

A comprehensive interview with parents is recommended to define symptomology and determine level of impairment. A well-established, semi-structured interview is the Schedule for Affective Disorders and Schizophrenia (SADS) (Kaufman et al., 1997). A clinical interview with the child, as well as behavioral observations in multiple settings, will also be beneficial. The Kiddie-SADS (K-SADS) was developed for school-aged children. The K-SADS- Present and Lifetime (K-SADS-PL) integrates interviews from the parent and child, as well as clinical observations, into summary ratings that are useful for diagnostic considerations.

Adaptive functioning is paramount when evaluating COS. Assessment across various realms will be vital to define impairment and where intervention should be implemented. Adaptive functioning can be assessed using questionnaires from parents, teachers, and self-reports. Specific areas should include functioning in school and domestic settings, with peer and family relationships, and self-care skills.

Cognitive functioning should be evaluated using standardized measures. Both verbal and nonverbal measures will be beneficial. Verbal components will provide the clinician with opportunities to assess thought processes using verbal means. Nonverbal assessments will be beneficial to assess underlying cognitive processes with reduced language- and communication-loading factors which have been recognized as possible deficits in children with COS (Caplan, 1994). Working memory, attention, and processing speed should also be evaluated. Deficits in these areas have been noted in the research literature but must be distinguished from more typical neurodevelopmental disorders (Reichenberg et al., 2010). Cognitive functioning may also help clinicians determine the child's level of expected functioning relative to their peers.

Language knowledge and communication skills should be assessed to determine levels of functioning in these areas. Evaluation of receptive and expressive language knowledge, as well as verbal comprehension, will help determine how the child is processing verbal information and their ability to express themselves to others.

Negative symptoms such as depression, catatonic behaviors, and avolition should be evaluated through observations, clinical interviews, and parent, teacher, and self-report questionnaires. Impairment in interpersonal functioning is often a result of symptoms as well. Personality factors and a sense of resiliency should be assessed through normative measures, while social skills should be assessed through questionnaires and behavioral observations. Anxiety and stress should be assessed in relation to impairment in the child's life. If warranted, suicide potential should also be evaluated.

Associated Impairments and Comorbid Diagnoses

Reserachers found that 81 of 82 children and adolescents in their study, who were diagnosed with COS, had a comorbid diagnosis (Ross, Heinlein, & Tregellas, 2006). ADHD was the

most common comorbid disorder with 84% of participants meeting the diagnostic criteria. Oppositional defiant disorder (43%), depression (30%), and separation anxiety disorder (25%) were also found to have high rates of comorbidity. Anxiety-related disorders such as OCD (24%), panic disorder (24%), and social phobia (17.7%) as well as eating disorders such as bulimia (4.9%), have also been found to have high rates of comorbidity with psychotic disorders (Cassano, Pini, Saettoni, Rucci, & Dell'Osso, 1998).

Not surprisingly, high rates of comorbidity (28%) exist between COS and ASD (Rapoport, Chavez, Greenstein, Addington, & Gogtay, 2009). In the DSM-II, behaviors associated with autism were classified as "childhood schizophrenic" (APA, 1968). Children met the diagnostic criteria for this disorder if they had "autistic, atypical, and withdrawn behavior" (p. 53). Although the authors of the DSM have separated these into two distinct disorders over time, overlapping symptoms still remain apparent.

Treatment

Identifying symptoms, as well as understanding the developmental course and prognosis, are important when working with children with COS. Identifying treatments that safely improve functioning in these children is equally important. Treatments for COS often include both pharmacological and psychotherapeutic interventions. Both routes may be equally important because of their differential effects on a child's functioning. Pharmacological interventions work to reduce positive and negative symptoms. Psychotherapeutic interventions enhance social skills, life skills, and provide family support.

Pharmacological Interventions

Antipsychotic medication was found to have a large intervention effect on overall functioning for individuals with AdOS in a recent meta-analysis (Stafford et al., 2015). Despite this, medication was found to have only a small effect in alleviating positive and negative symptoms while enhancing psychosocial functioning. Analyses differentiating treatment effects for specific medications are still in its infancy and should be further researched. These researchers noted that many head-to-head medication studies included in their analysis were underpowered or had low quality, making any indication of comparisons unreliable at this time.

Pharmacological interventions are similar to those given to individuals with AOS and often include typical and atypical antipsychotics. Their effectiveness and common significant side effects are presented below:

Typical Antipsychotics

Typical antipsychotics, or first-generation antipsychotics, were developed to reduce symptoms of psychosis. Medications include chlorpromazine, droperidol, haloperidol, and many others. Typical antipsychotics have been extensively studied in adult populations, but their use and effectiveness with children is limited.

An early study examined the effects of haloperidol in a randomized double-blind, placebo-controlled crossover study with children with COS (Spencer, Kafantaris, Padron-Gayol, Rosenberg, & Campbell, 1992). Researchers found haloperidol to be superior to placebo on several measures of symptom severity. Using the same treatment sample in an expanded study, researchers found haloperidol to be effective in improving daily functioning (Spencer & Campbell, 1994). Its effectiveness was positively related to the participant's age and intellectual functioning, while inversely related to the onset of symptoms. More specifically, older children, those with higher intellectual abilities, and those with later-onset symptoms showed a greater reduction of symptoms when taking haloperidol compared to placebo.

Typical antipsychotics have shown some promise but, similar with adult populations, have been criticized. Typical antipsychotics tend to only alleviate positive symptoms of schizophrenia and often result in significant adverse side effects.

Positive symptoms may be more readily apparent, but negative symptoms (catatonia, avolition, depression, etc.) often contribute to long-lasting impairment. Significant side effects of typical antipsychotics are also deterrents. Extrapyramidal symptoms (EPSs) may present more frequently in children, when compared to adults, and may contribute to significant and lasting impairments (Kumra et al., 1998).

Atypical Antipsychotics

Atypical antipsychotics, or second-generation antipsychotics, are prescribed to alleviate positive and negative symptoms of schizophrenia. They are commonly associated with less severe side effects compared to traditional antipsychotics. Because of this, they are often considered a first-line pharmacological treatment in children and adults. Atypical antipsychotics include clozapine, risperidone, and olanzapine, among others.

Clozapine Clozapine was the first marketed atypical antipsychotic and has been one of the most researched medications for children with COS. It is often considered a first-line medication in adults, but due to some of the serious adverse effects of this medication, caution is warranted with children.

In an early double-blind study comparing the effects of clozapine to haloperidol, evidence supported the effectiveness of clozapine in children and adolescents with COS (Kumra et al., 1996). Turetz et al. (1997) studied clozapine in an open-label, 16-week trial with 11 participants with COS who were resistant to at least two treatments of either typical or atypical antipsychotics. At a dosage of 227.3 + or −34 mgs per day, there was a reduction of psychotic symptoms on all measures at the conclusion of the study. Peak efficacy of symptom reduction was found to be within 6 and 8 weeks of beginning clozapine.

The effectiveness of clozapine versus olanzapine was studied in a double-blind, randomized, 8-week controlled trial with a 2-year open-label follow-up (Shaw et al., 2006). Participants aged 7–16, who showed resistant to at least two other forms of antipsychotics, were randomly assigned to either a clozapine or olanzapine treatment group. Participants in the clozapine treatment group showed significant improvement in all outcome measures (reduction in psychotic symptoms), whereas participants in the olanzapine treatment group showed less consistent improvement. Moderate to large treatment effects favored the clozapine treatment group, compared to an antipsychotic medication-free baseline phase, but these effects were only significant in alleviating negative symptoms.

The use of clozapine to reduce antipsychotic symptoms was further studied using either a double-blind or open-label procedure with 54 participants (Sporn et al., 2007). A reduction of symptoms was found at 6 weeks of clozapine use, and functional improvements were apparent at the 2- to 6-year follow-up.

Significant side effects associated to clozapine have been identified. Tachycardia, hypertension, cardiac and lipid abnormalities, agranulocytosis, seizures, enuresis, and other side effects may be detrimental to a child's development and result in lasting impairment (Kumra et al., 1996; Shaw et al., 2006). Clozapine is also considered a more expensive treatment route, due to its high costs and the required weekly blood monitoring of white cell count due to the possibility of agranulocytosis (American Academy of Child and Adolescent Psychiatry [AACAP], 2001; Dulmus & Smyth, 2000). These concerns have resulted in clozapine being used cautiously, when other medications have proven ineffective.

Risperidone Risperidone is another atypical antipsychotic commonly used to treat positive and negative symptoms associated with COS. In one double-blind study, participants were randomly assigned to a risperidone, olanzapine, or haloperidol treatment group to assess efficacy and tolerability over an 8-week trial period (Sikich, Hamer, Bashford, Sheitman, & Lieberman, 2004). All medications were found to have a similar reduction of psychotic symptoms, but the risperidone group reported less adverse side effects over time.

Haas et al. (2009) also studied risperidone in alleviating positive and negative symptoms, along with its negative side effects. In a 6-week double-blind, placebo-controlled study across 160 participants, risperidone was found to be well tolerated compared to placebo. Adverse side effects such as EPSs were more common in the treatment group, yet the severity of these was reported to be low.

Alleviation of positive and negative symptoms, along with its tolerable side effects, has resulted in risperidone being considered the more appropriate treatment for children with COS compared to clozapine and typical antipsychotics. Although promising, research is still limited with risperidone, and should be studied further in children and adolescents.

Olanzapine Olanzapine, another atypical antipsychotic, has also shown promise alleviating psychotic symptoms associated with COS. In the aforementioned Sikich et al. (2004) study, olanzapine showed a similar effectiveness in reducing psychotic symptoms compared to risperidone and haloperidol. Moreover, in a 1-year, open-label trial study of olanzapine, researchers found a reduction in positive symptoms after 6 weeks of medication use and improvement in negative symptoms after 1 year (Ross, Novins, Farley, & Adler, 2003).

Like risperidone, olanzapine is not without negative side effects. Olanzapine may produce similar negative side effects as other atypical antipsychotics, as well as increased appetite, weight gain, gastrointestinal symptoms, and metabolic abnormalities (Remschmidt, Hennghausen, Clement, Heiser, & Schulz, 2000; Toren, Ratner, Laor, & Weizman, 2004). Future research should also be conducted to further understand this medication in children and adolescents.

Other Atypical Antipsychotics Other atypical antipsychotics, including quetiapine, ziprasidone, aripiprazole, and molindone, have yet to be studied extensively in children and adolescents. Initial research has led to promising results in alleviating symptoms associated with COS, but more controlled studies are needed (Lindsey, Kaplan, & Koliatsos, 2003; McConville et al., 2003). Treatment efficacy, tolerability of side effects, and comparability with other typical and atypical antipsychotics should be evaluated.

Adverse Side Effects

Adverse side effects should be considered with any pharmacological intervention. EPSs including dystonias, Parkinsonisms, akathisia, and tardive and withdrawal dyskinesias have been associated with both typical and atypical antipsychotics. EPSs can occur in low or cumulative doses and/or long exposure to traditional and/or atypical antipsychotics. However, in a comparative study of effectiveness and tolerability, EPSs were found to be more frequent and severe in participants treated with haloperidol versus risperidone or olanzapine (Sikich et al., 2004).

Weight gain is consistently reported when using atypical antipsychotics. Sikich et al. (2004) found that participants gained weight in all treatment groups, yet atypical antipsychotic use (olanzapine and risperidone) resulted in a significantly faster rate of weight gain compared to the haloperidol group. Increased weight gain can lead to obesity and/or metabolic dysfunction such as alterations in glycemia, hyperprolactinemia, and hepatotoxicity. Increased weight gain should also be considered within a developmental context, as it may contribute to negative internalizing symptoms associated with self-esteem and self-worth.

Neuroleptic malignant syndrome (NMS) is a potentially life-threatening side effect of typical and atypical antipsychotics. NMS is characterized by high fever, sweating, unstable blood pressure, muscular rigidity, autonomic instability, and altered mental status. NMS can lead to a loss of consciousness and/or death. The severity of NMS often leads to an immediate discontinuation of an antipsychotic medication and treatment with electrolyte infusions and hydration.

Seizures have been associated with antipsychotic medications but may be more common in atypical antipsychotics (Lertxundi et al., 2013). Increased risks are often associated with clozap-

ine use, but research has also found risperidone and quetiapine to have elevated risks of epileptic seizures (Centorrino et al., 2002). Because of this, children with a history of seizures should receive electroencephalogram (EEG) monitoring when using antipsychotic medications.

Hematological side effects are also associated with clozapine use (Hummer, Kurz, Barnas, Saria, & Fleischhakker, 1994). White blood cell count should be consistently monitored throughout treatment. Other adverse side effects such as cardiovascular effects have been extensively studied in adults but are rarely reported in children and adolescents. Albeit rare, these side effects should be monitored consistently throughout pharmacological treatments.

Non-pharmacological Treatment Interventions

Pharmacological therapies such as typical and atypical antipsychotics are often used to alleviate positive and negative symptoms associated with COS. Psychological treatments should also be used in conjunction with any medication regimen. Combining family interventions with individual cognitive behavioral therapy (CBT) had strong intervention effects for young adults on relapse of symptoms in the aforementioned meta-analysis (Stafford et al., 2015). More specifically, the duration between psychotic symptoms was significantly increased when combining family intervention with individual CBT. While no psychological treatments for children or adolescents were included in this meta-analysis, promising results from young adult studies may be extended downward with some caution.

Despite this, psychotherapeutic treatments included in this meta-analysis were not found to significantly decrease psychotic symptoms or alleviate depressive symptoms. The researchers noted that research in this area was limited though. As with all meta-analyses, due to the various conditions and sample characteristics of included research studies, it is difficult to draw affirmative conclusions of treatments effects.

Cognitive strategies, family interventions, educational interventions, and environmental manipulation are recommended psychological treatments (Gonthier & Lyon, 2004). Support for key stakeholders (family, teachers, etc.) should also be considered.

Cognitive therapy may include social skills support, life skills training, relapse prevention, and developing self-monitoring strategies (AACAP, 2001). As noted previously, children with COS may experience decreased social functioning, resulting in social withdrawal and isolation. Children may also demonstrate a limited use of self-care skills and demonstrate difficulty focusing and attending to information. Enhancing previously developed skills, as well as teaching underdeveloped skills, will be paramount. Social skills training may include developing communication skills, assertiveness, and self-advocacy. Life skills may include formulating coping strategies, vocational training, and utilizing self-care skills such as hygiene maintenance, cooking, and money management (Dulmus & Smyth, 2000). Self-monitoring strategies should focus on assisting the child or adolescent to recognize both positive and negative symptoms in various situations, and how to seek support if needed.

Presenting information on COS, the typical developmental course of the disorder, and prognosis can help the family understand COS. Developing coping strategies for family members, such as problem-solving and communication skills, will be critical. Defining and understanding family characteristics that promote, or hinder, the child's emotional well-being should also be a focus. Reducing expressed emotion (overly critical, hostile, intrusive, and highly expressive emotions) and environmental stressors are essential strategies used to reduce the risk of relapse of prodromal and acute phase symptoms (Lopez et al., 2004).

Symptoms of COS may result in a negative impact on the child's academic, behavioral, and/or social relationships at school. Specific interventions or accommodations may be provided to support the child in educational environments. The child's educational team should consider

support through special education services under the Individuals with Disabilities Education Improvement Act (IDEIA) or specific accommodations under the Americans with Disabilities Act (ADA), Section 504. Interventions may include social skills intervention, academic remediation instruction, or social-emotional support. Developing functional life skills and providing vocational training and support will be important in middle school and high school. Accommodations may include smaller class sizes, reduced distractions, study skills courses, and specific teachers with experience working with children with COS. Modification of assignments with an emphasis on the mastery of concepts may be beneficial to reduce stress. Short/specific direct instruction due to poor attention abilities and low frustration tolerance may also be warranted.

Environmental manipulation may also reduce the severity or occurrence of symptoms. These are often incorporated into family therapy and educational interventions. Creating consistent, viable structures within family and school settings will create a sense of routine for all children. For children with COS, this may result in a decreased likelihood and severity of symptoms. If symptoms are severe enough to warrant hospitalization, environmental manipulation is essential for improvement and recovery.

Conclusion

Childhood-onset schizophrenia is a rare psychological disorder. Estimates are that 1 in 10,000 children will demonstrate symptoms that meet the diagnostic criteria. Positive symptoms such as delusions, hallucinations, disorganized speech, and disorganized behaviors are often most readily present. Negative symptoms such as a lack of emotional expressivity, catatonia, avolition, and depression may be more difficult to detect. Family, school, and social functioning are most often negatively affected. To meet the full criteria for schizophrenia, symptoms must also persist for at least 6 months, although children may meet the diagnostic criteria for other schizophrenia spectrum disorders with less persistent symptoms.

There is an abundance of genetic, neuroimaging, and environmental studies examining COS. It is safe to assume that this disorder is not due to a specific gene mutation, brain abnormality, personality type, or environmental stressor. Rather, COS is likely the result of a constellation of these. Due to the complex presentation of symptoms, assessment should be comprehensive. Integrating information from clinical interviews and observations, with data of intellectual and neuropsychological abilities, language knowledge, behavioral skills, and adaptive and emotional functioning will be paramount.

The developmental course of COS is often described in four distinct, yet fluid, phases. The prodromal phase is characterized by functional deterioration and onset of psychotic symptoms. Social withdrawal and isolation, as well as decreased use of self-care skills, may result. The acute phase consists of a predominance of positive symptoms. The severity of symptoms may result in a deterioration of cognitive and social functioning, and may require hospitalization for diagnosis and treatment. In the recuperative or recovery phase, children are often able to function outside inpatient or residential settings due to a decrease in positive symptoms. Poor attention, bizarre behaviors, and negative emotions can significantly impair functioning. Children in this phase may also experience internalizing symptoms, demonstrate a flat affect and apathy, and be socially withdrawn. In the residual phase, positive and negative symptoms diminish from previous levels. Children have improved functioning, but continue to be impaired relative to their premorbid functioning.

Children may fluidly move throughout these phases or cycle back to prodromal or acute phases. A relapse is considered a return to, or worsening of, psychotic symptoms (a return to a predominance of psychotic symptoms after improved functioning). It is paramount that adequate treatment be provided at these times to decrease the likelihood of relapses.

Treatment of COS, like the evaluation of the disorder, should be multidimensional. Alleviating symptoms and reducing adverse side effects

should be considered within the developmental context of the child when using any pharmacological intervention. Atypical antipsychotics such as risperidone and olanzapine may be effective in relieving both positive and negative symptoms associated with COS. These are often considered first-line pharmacological treatments for COS due to their effectiveness and tolerability. Clozapine has been extensively studied but may present with more severe side effects relative to other atypical antipsychotics. Typical antipsychotics such as haloperidol may be beneficial in alleviating positive symptoms, but may result in significant side effects.

Psychotherapeutic interventions include cognitive, family, and educational interventions. These are used to improve the long-term outcomes of children with COS. Providing information and support to the family and school may also reduce the likelihood and severity of symptoms through environmental manipulation.

Research examining the long-term outcome of children with COS has found that most children are moderately to severely impaired into adulthood. Children often meet the diagnostic criteria of schizophrenia as adults or continue to demonstrate symptoms. These adults tend to have difficulty developing and maintaining social and romantic relationships. Obtaining and maintaining employment may also be difficult. The majority of these adults need continual pharmacological and psychological treatment along with educational and vocational support. Due to both positive and negative symptoms, children, adolescents, and adults are also at an increased risk for suicide.

COS represents a more severe form of schizophrenia. There is a high likelihood of symptoms persisting into adulthood, despite effective treatments. Objectives for researcher and clinicians should include increasing the understanding of COS, developing more refined diagnostic procedures, and improve functioning through both pharmacological and psychotherapeutic interventions. The importance of early identification and treatment cannot be understated. Future research is paramount to promote positive, long-term outcomes for this population.

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Part IV

Internalizing Disorders: Mood Disorders

Bipolar Disorder in Children

6

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History of Bipolar Disorder

Although childhood bipolar disorder was not formally identified as a diagnostic entity until the twentieth century, careful analysis of the adult presentation paved the way. The origins of bipolar disorder date back to ancient Greece (Glovinsky, 2002), with depression and mania being among the first documented mental illnesses in adults (Angst & Marneros, 2001). The Greek physician Aretaeus of Cappadocia did not define bipolar disorder as being two separate entities of melancholia and mania, instead suggesting that mania was a worsened state of melancholia (i.e., severe depression) (Angst & Marneros, 2001). From here, the literature does not reflect a clear connection between depression (i.e., melancholia) and mania until the seventeenth century, when clinicians such as Theophile Bonet and Willis began describing these mental states under the broad category of mood disorders (Altschule, 1986). In the middle of the nineteenth century, the French psychiatrist Jean-Pierre Falret further clarified that bipolar disorder was cyclical in nature (i.e., *folie circulaire*) with depressive and manic states clearly separated by symptom-free intervals (Glovinsky, 2002). This revolutionized the conceptualization of bipolar disorder by not only tethering two drastically different mood presentations together but also by directing the scientific community to focus on the temporal aspects of mood. While controversy erupted over diagnostic details even then, many leading clinicians agreed that the prognosis of bipolar disorder was poor (Marneros, 2009). The turn of the twentieth century marked the general acceptance of bipolar disorder as a clinical diagnosis, particularly with the added support of the astute observations of German psychiatrist Emil Kraepelin. Kraepelin studied the natural course of the untreated disorder and found it to be punctuated by relatively symptom-free intervals. Based on these observations, he distinguished the disorder from *dementia praecox* (i.e., *schizophrenia*) and coined the term "manic-depressive insanity (MDI)." Kraepelin advocated for the absorption of bipolar disorder into the category of MDI, which included unipolar depression (i.e., single episode and recurring depression) and mood-driven psychosis (Angst & Sellaro, 2000). He suggested that individuals with this diagnosis had an episodic course and a more benign functional outcome, relative to patients with *dementia praecox*, which he conceptualized as having a progressive deteriorating course. Kraepelin also alluded to the idea that bipolar disorder could occur in prepubertal children, though described this as a rare entity and limits discussion to juvenile mania (Kraepelin, 1921).

Kraepelin' s unification of mood disorders under one heading (i.e., MDI) halted individual exploration of mania, depression, and bipolar

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disorder, as researchers placed little emphasis on differentiating between the three mental states (Pollock, 1931). It took the publication of two seminal papers approximately 50 years later to revisit the diagnostic conceptualization of bipolar disorder, coined the "rebirth" of bipolar disorder in 1966 (Marneros & Angst, 2000). The pioneering authors, Angst (1966) and Perris and d'Elia (1966), presented information contradicting the unification model of mood disorders and instead highlighted key differences between unipolar depression and bipolar disorder. They observed that bipolar disorder was represented equally among the sexes, while unipolar depression was more prominent in the female gender; that bipolar disorder was not homogenous, while unipolar depression had a much more classic presentation with similar presenting symptoms; and that genetic factors are critically involved in both disorders. Ultimately, this work facilitated increased interest in the origins and pathophysiology of bipolar disorder.

Up until the twentieth century, little attention was devoted to bipolar disorder in children. In the 1920s, child psychiatrists began searching for bipolar disorder in their patients, concluding that, overall, cases were rare, with the majority presenting in adolescence (Angst & Sellaro, 2000; Glovinsky, 2002). Many psychiatrists noted the difficulty with drawing a clear diagnostic distinction between mania and attention-deficit hyperactivity disorder (ADHD), given symptomatic overlap in excitability profiles, poor stress tolerance, and irritability. This controversy ignited one of the most heated issues in child and adolescent psychiatry to date, which would reach its potential later in the century (Glovinsky, 2002). In 1960, Anthony and Scott conducted a systematic review of the literature, which reiterated that bipolar disorder was rare in children (Anthony & Scott, 1960). This topic was revisited in the 1970s, as interest in understanding and identifying bipolar disorder in children peaked (Glovinsky, 2002). Researchers and clinicians began refining the diagnostic criteria of childhood bipolar disorder, with Weinberg and Brumback creating criteria to diagnose mania in children. Ultimately, their work would serve as the basis for the DSM-III bipolar disorder criteria (not specific to children), where mania was defined by symptoms of euphoric or irritable mood, hyperactive behavior, and flight of ideas included under the manic presentation. Since the early 1980s, DSM criteria have specified that adult criteria can be used to diagnose mania in children, with appropriate modifications for age and developmental stage (Weller, Weller, & Fristad, 1995). As a result, by the 1980s, the research community began to accept the notion that children could present with bipolar symptoms, though the expectation persisted that incidence would be rare. Subtleties in childhood presentation of mania as compared to adult onset included intrinsic hyperactivity; the absence of discrete, specific episodes (i.e., more of a blended symptom presentation); and a high likelihood of irritability and emotional lability versus euphoria (Kleist, 1912). Children in the depressive phase were conceptualized as being similar to children with unipolar depression in that they were agitated and irritable, with decreased stress tolerance (Leonhard, 1957).

Bipolar disorder in children remains a controversial topic in the field of child and adolescent psychiatry. Comorbidity with other mental illnesses, diagnostic criteria, and consequent treatment are among the issues smoldering in the international community. The Word Health Organization (WHO) US/UK study of bipolar disorder and schizophrenia demonstrated real challenges in the accurate diagnosis of childhood bipolar disorder, through highlighting different diagnoses made by each respective country based on the same symptom presentation (Cooper, Kendell, Gurland, Sartorius, & Farkas, 1969). This discovery in the late 1960s/early 1970s facilitated a global effort to standardize diagnosis through operationally defining criteria. Structured and semi-structured interviews were consequently developed with bolstered reliance on standardized criteria for diagnosis (American Psychiatric Association [APA], 1980; Spitzer, Endicott, & Williams, 1979). Despite multiple efforts, however, recent studies perpetuate prior findings, showing that clinicians in the United States continue to have a lower threshold for

diagnosing childhood bipolar disorder relative to other countries (Dubicka, Carlson, Vail, & Harrington, 2008; Stringaris & Youngstrom, 2014). In fact, a recent study completed by James and colleagues suggests that pediatric bipolar disorder is more commonly diagnosed in inpatient settings in the United States than in England, with discharge rates 12.5 times higher in the United States compared to England (James et al., 2014). This finding mirrors results of other inpatient surveys conducted throughout the world (where percentage rates of total hospital admissions for pediatric bipolar disorder are given as a percentage of all pediatric psychiatric hospitalizations), with Denmark ranging from 1.2% to 1.7% (Thomsen, Moller, Dehlholm, & Brask, 1992) and 2.5–4.2% in India (Alexander & Raghavan, 1997; Reddy, Girimaji, & Srinath, 1997), compared with 27.8% in the United States (James et al., 2014). There are multiple possibilities for this diagnostic discrepancy in the global community, with the leading explanation pointing to differences in diagnostic criteria used. Other countries, specifically England, use the ICD-10 versus the more commonly used DSM in the United States. The ICD-10 requires more than one manic episode to meet criteria for bipolar disorder, whereas the DSM-IV-TR and DSM-V require only one such episode. Additional research investigating this putative cultural divide is needed.

Diagnostic Criteria (DSM-V)

The DSM-V was released in May 2013, with few changes made to the bipolar disorder diagnostic criteria from the DSM-IV-TR (APA, 2013) (Table 6.1). The criteria are largely the same for children as adults, though there are a few notable differences in symptom presentation. Specifically, depression in children may not present as anhedonia and depressed mood but may present more as irritability. A depressive episode occurs when an individual endorses either a depressed mood or anhedonia, with disturbances in sleeping and eating habits, psychomotor slowing or agitation, feelings of guilt, decreased concentration, lack of energy, and suicidal ideation for at least 1 week. These symptoms usually result in some level of dysfunction and cannot be attributed to a medical etiology or exogenous substance (APA, 2000). A manic episode, on the other hand, is identified when individuals have elevated, expansive, or irritable mood that lasts for at least 1 week or, if less than a week, requires hospitalization (APA, 2000). Presenting symptoms may include grandiosity, decreased need for sleep, pressured speech, flight of ideas, distractibility, and increased goal directed activity, along with excessive participation in pleasurable activities (i.e., hypersexuality, spending large quantities of money, etc.). When an individual presents with an elevated/expansive mood, at least three or more of these symptoms must be present to meet diagnostic criteria, while four or more symptoms must be present if with irritable mood. Similar to depression, this constellation of symptoms must cause impairment in functioning without being attributable to a medical etiology or substance. The criteria are the same for a hypomanic episode, though the duration of time is decreased from 7 to 4 days. Changes were made to the DSM-V criteria for a mixed episode in that full criterion for both manic and depressive episodes for greater than 1 week is no longer necessary, being replaced by a mixed features specifier. This specifier is applied when individuals meet full criteria for a depressive episode and at least three symptoms of a manic episode or when individuals meet full criteria for a manic episode and at least three symptoms of a depressive episode. Ultimately, this change may increase the frequency of those who meet mixed episode criteria, including children.

The DSM-V, much like the DSM-IV-TR, includes bipolar I disorder, bipolar II disorder, and cyclothymia. In order to receive a bipolar I disorder diagnosis, individuals must have experienced at least one manic episode in their lifetime. To meet criteria for bipolar II disorder, individuals must experience at least one depressive episode and at least one hypomanic episode. Of note, if these individuals have experienced manic symptoms for seven or more days, they meet criteria for a manic episode, which is mutu-

Table 6.1 Selected DSM-5 mood disorders and the episodes required for diagnosis

Diagnosis	Criteria/time	Symptoms/presentation	Manic episode	Hypomanic episode	Major depressive episode
Bipolar I disorder	≥3 symptoms for ≥1 week (or any duration if hospitalized)	Mania: grandiosity, decreased need for sleep, pressured speech, racing thoughts, distractibility, increased goal-directed activity, risky behavior	Yes	Common	Common
Bipolar II disorder	≥3 symptoms for ≥4 days	Hypomania: grandiosity, decreased need for sleep, pressured speech, racing thoughts, distractibility, increased goal-directed activity, risky behavior *without* psychosis or hospitalization	No	Yes	Yes
Cyclothymic disorder	Symptoms present for at least half the time with an absence of symptoms ≤2 months during this time; ≥2 years in adults and ≥1 year in children	Multiple hypomanic and depressive symptoms that do not meet criteria for a hypomanic or major depressive episode	No	No	No
Disruptive mood dysregulation disorder	Specific to children up to 12 years of age. Irritable mood present most of the day, nearly every day. Temper outbursts ≥3 times per week for ≥1 year in ≥2 settings	Chronic, severe, persistent irritability manifesting in temper outbursts and severe irritability that is non-episodic	No	No	No
Unipolar major depressive disorder	≥5 symptoms nearly every day for ≥2 weeks *Can also meet criteria for persistent depressive disorder	Sadness/anhedonia, weight change, sleep disturbance, psychomotor agitation/retardation, low energy, guilt, poor concentration, and suicidality	No	No	Yes
Persistent depressive disorder (dysthymia)	≥3 symptoms more days than not for ≥2 years in adults and ≥1 year in children	Sadness, weight change, sleep disturbance, low energy, low self-esteem, poor concentration, hopelessness *Anhedonia, psychomotor agitation/retardation, guilt, and suicidality are *not* part of the criteria	No	No	No

Other specified bipolar and related disorder (replaced BP-NOS)	Does not meet criteria of any other bipolar or depressive disorder and not due to substances/medications or a medical condition	The specific reasons for not meeting criteria are specified and include: Major depressive episodes and short-duration hypomanic episodes Major depressive episodes and hypomanic episodes with insufficient symptoms Hypomanic episode without prior major depressive episode Short-duration cyclothymia	No	Maybe *If yes, must not meet criteria for a major depressive or manic episode	Maybe *If yes, must not meet criteria for a hypomanic or manic episode
Unspecified bipolar and related disorder (replaced BP-NOS)	Used when clinicians choose *not* to specify the reason that criteria are not met, including when there is insufficient information (e.g., emergency room settings)	Symptoms characteristic of a bipolar and related disorder that cause distress/impairment, though do not meet full criteria for unspecified reasons	No	No	No

ally exclusive with a bipolar II disorder diagnosis (APA, 2013). In order to distinguish an endogenous vs. exogenously induced symptom presentation, there are diagnostic distinctions made for substance-induced, medication-induced, and bipolar disorder due to a medical condition (APA, 2013).

Symptom Presentation

While the DSM-V has certainly allowed for increased diagnostic latitude, many find making the diagnosis of bipolar disorder in children difficult. This has been attributed to multiple reasons, including, but not limited to, the increased frequency of mixed episodes in children (Findling et al., 2001; Pavuluri, Birmaher, & Naylor, 2005), the unreliability of self-report (Birmaher et al., 2006), the ambiguity of symptoms, the difficulty meeting symptom duration criterion (Pavuluri et al., 2005; Sala, Axelson, & Birmaher, 2009), and the difficulty in making clear distinctions between clinically significant symptoms and developmentally normative behaviors (see Sect. "Symptom Presentation" for further discussion) (Carlson, 2005; Luby & Belden, 2006).

Briefly, the symptom presentation in children mirrors that of adults, with a few notable differences (Table 6.2). Depression in children may not manifest as depressed mood and anhedonia but may present as irritability. Therefore, along with possible alterations in sleeping and eating, psychomotor slowing, feelings of guilt, decreased concentration and energy, and/or suicidal ideation lasting for at least 1 week, children with bipolar disorder who are in a depressive phase may present as agitated and edgy. A manic episode, on the other hand, occurs when children endorse an elevated, expansive, or irritable mood that lasts for at least 1 week or, if less than a week, requires hospitalization. Presenting symptoms may include grandiosity, decreased need for sleep, pressured speech, flight of ideas, distractibility, and increased goal-directed activity, along with excessive participation in pleasurable activities. The most common symptoms, however, are increased energy, distractibility, and pressured speech (Kowatch, Youngstrom, Danielyan, & Findling, 2005). Similar to depression, symptoms must ultimately cause impairment in functioning without being attributable to an extrinsic etiology (i.e., medications or substances). Youth with bipolar disorder typically experience significant impairment in academic functioning and social interactions and may have lower self-esteem (Geller, Bolhofner et al., 2000). Furthermore, although bipolar disorder is episodic in nature, it can present with or without inter-episode recovery. This may make diagnosis more difficult, as episodes in children can be more amorphous with less clinically defined endpoints. Finally, it is important to point out that irritability is included in both manic and depressive episodes, which elevates this symptom to critical importance in this population.

When evaluating irritability in children and adolescents, it is important to recognize that this symptom is not pathognomonic to bipolar disorder but is rather ubiquitous in childhood mental illness (Biederman et al., 2004; Geller et al., 2002; Leibenluft, 2011; Leibenluft, Charney, Towbin, Bhangoo, & Pine, 2003). Furthermore, this symp-

Table 6.2 Differences between childhood and adult bipolar disorder

	Childhood bipolar disorder	Adult bipolar disorder
Onset	Prepubertal, early adolescent	Late adolescent, early 20s
Symptom duration	Longer symptomatic duration; decreased frequency of discrete, specific episodes	Episodic
Mixed episodes	Common	Less common
Family history	Common	Less common, though not uncommon
ADHD comorbidity	60–90%	Less common
Manic symptoms	Irritability	Grandiosity, euphoria
Depressive symptoms	Irritability	Depressed mood, anhedonia
Psychotic symptoms	Less common	More common

tom, if not impairing function, is a component of normal childhood development. Of note, irritability is a presenting symptom in at least six childhood disorders including major depressive disorder, generalized anxiety disorder, posttraumatic stress disorder, ADHD, ODD, and conduct disorder (Mick, Spencer, Wozniak, & Biederman, 2005; Stringaris, 2011). Researchers have found that the level of irritability in children and adolescents with bipolar disorder is severe, persistent, and highly disabling, often manifesting in violence (Biederman et al., 2004). In fact, a meta-analysis completed by Kowatch and colleagues found that 81% of children with bipolar disorder experienced irritability (Youngstrom et al., 2005). While the DSM-V does not explicitly define irritability, other definitions—mostly diagnostic scales—exist in the literature. For example, the Schedule for Affective Disorders and Schizophrenia for School-Age Children (K-SADS) defines irritability as "anger, crankiness, bad or short tempered, resentment or annoyance, touchy or easily annoyed" (Kaufman et al., 1997), while the Diagnostic Interview Schedule for Children (DISC) operationalizes it as "cranky, angry toward people you had no reason to, talking back, or temper tantrums" (Shaffer, Fisher, Lucas, Dulcan, & Schwab-Stone, 2000). Interestingly, as outlined in the DSM-V, while mania criteria can be met with three additional symptoms when a child presents with elevated mood, at least four additional symptoms must be present when only irritable mood is present. Some researchers argue that these rigid criteria for mania are the rate-limiting step in offering correct diagnoses to children with bipolar disorder who may benefit from early intervention (APA, 2000; Axelson et al., 2006; Birmaher et al., 2006; Findling et al., 2001; Staton, Volness, & Beatty, 2008). Just like any clinical symptom, however, there is a certain amount of subjectivity, making it difficult to identify irritability as a symptom associated with manic presentation with a high degree of certainty.

As symptoms such as irritability and emotional lability are observed in multiple disorders of childhood, it is critical to adhere to the operational definitions of mania and hypomania provided in the DSM-V. Strict adherence will foster increased diagnostic validity that may bolster clinicians' confidence that they are indeed treating the correct diagnosis. In particular, it is prudent to pay close attention to the pattern and duration of bipolar symptoms and how these symptoms affect sleep, motor/physical activity, and changes in cognitive function across multiple domains. Through such vigilance, clinicians can feel more confident that they are targeting symptoms specific to bipolar disorder and not incorrectly diagnosing another childhood mental illness.

Associated Impairments and Comorbidities

Children with bipolar disorder tend to have impairments in cognitive, emotional, social, and academic performance, with a higher likelihood compared to the general population of legal problems and suicidal or self-injurious behavior (McClellan, Kowatch, & Findling, 2007). The clinical picture of childhood bipolar disorder is complicated by the high prevalence of comorbid psychiatric disorders. Differential diagnosis may be difficult given that bipolar disorder and other diagnoses include symptoms that are common in children with and without psychiatric illness. Clinicians working with children should, therefore, be aware of co-occurring diagnoses, as some distinctions are more challenging than others.

ADHD

It has been shown that 60–90% of children and adolescents with bipolar disorder suffer from comorbid ADHD. This can be partially attributed to the large overlap in diagnostic criteria between each diagnosis, including symptoms of distractibility, hyperactivity, and pressured speech. In fact, even when these overlapping symptoms are removed from the diagnostic count, approximately 89% of youth with bipolar disorder continue to meet full criteria for ADHD (Doerfler, Connor, & Toscano, 2011). When distinguishing bipolar disorder from ADHD, it is important to

note that children with bipolar disorder have subtle episodic fluctuations and distinct symptoms of mania (Youngstrom, Birmaher, & Findling, 2008). For example, while children with bipolar disorder may have decreased need for sleep, children with ADHD can present with insomnia, although their need for sleep is unchanged (Goldstein, 2012). Furthermore, understanding the specific symptoms of mania is critical, as children with bipolar disorder may present with grandiosity and hypersexuality, while these symptoms are not typical for ADHD. Recent research has identified important distinctions between children with bipolar disorder compared to youth with ADHD. Children with bipolar disorder were found to exhibit more reactive and verbal aggression along with more depressive symptoms than their ADHD counterparts (Doerfler et al., 2011). From a neurocognitive standpoint, children with ADHD demonstrate more executive function deficits than youth with bipolar spectrum disorders (Rucklidge, 2006). With regard to differences in brain structure and function, ADHD has been associated with decreased caudate and putamen volumes, while children with bipolar disorder tend to have larger caudate, putamen, and globus pallidus volumes, which was not accounted for by exposure to stimulant, antipsychotic, or mood stabilizer medication (Liu et al., 2011). From a global standpoint, childhood bipolar disorder can be conceptualized as an emotionally driven illness associated with a reward/affect system, while ADHD can be viewed as a cognitively driven, impulsive illness associated with behavior dysregulation (Singh, Ketter, & Chang, 2014).

Conduct Disorder

Rates of comorbid conduct disorder range from 48% to 69% among pediatric bipolar disorder cases. Biederman and colleagues found that the two common characteristics resembling mania also found in youth with conduct disorder are physical restlessness and poor judgment (Biederman, Faraone, Chu, & Wozniak, 1999). Other researchers have found evidence for overlapping symptoms of irritability, hostility, and impulsivity (Kim & Miklowitz, 2002). While irritability has been shown to be a presenting symptom in multiple child and adolescent disorders, irritability in bipolar and conduct disorders is often severe and violent (Wozniak et al., 1995). Behaviorally, children with either diagnosis can demonstrate inappropriate sexual behavior and disinhibited social interactions and display poor judgment such as stealing their parents' money and credit cards. Bowring and Kovacs argue that conduct disorder has a lengthy prodromal period with progression from less to more severe rule breaking, whereas mania is associated with a more abrupt onset of impulsive behavior (Bowring & Kovacs, 1992). However, they and others acknowledge the difficulty of identifying mania when comorbid conditions are present, as the temporal divisions are not always clear.

Anxiety Disorders

Children and adolescents with bipolar disorder have higher than expected rates of panic, generalized anxiety, and separation anxiety disorder (Castilla-Puentes, Sala, Ng, Galvez, & Camacho, 2013). In fact, lifetime prevalence of panic disorder was found to be approximately 21% among those with bipolar disorder compared with 0.8% in typically developing controls. Interestingly, patients with bipolar disorder and comorbid anxiety were reported to have higher rates of alcohol abuse and suicidal tendencies, compared to bipolar patients without comorbid anxiety. In contrast to the episodic nature of bipolar disorder, children with generalized anxiety disorder (i.e., without bipolar disorder) tend to have chronic symptoms such as persistent irritability, restlessness, and impaired concentration (Goldstein, 2012). In order for such symptoms to be considered symptoms of bipolar disorder, they must be specifically heightened from baseline during a mood episode (Goldstein, 2012).

Diagnostic Evolution: Severe Mood Dysregulation and Disruptive Mood Dysregulation Disorder

In an effort to differentiate broad and narrow bipolar phenotypes, Leibenluft and colleagues developed a new category outside of DSM called "severe mood dysregulation" (SMD) in 2003 (Leibenluft et al., 2003). Briefly, the more narrowly defined bipolar phenotype requires episodicity and elated mood/grandiosity in order to meet criteria, while the broad or liberal position is inclusive of children whose mood is largely irritable, lacking clear periods of grandiosity/elated mood. In order to meet criteria for SMD, children must present with severe episodes of rage in response to negative emotional stimuli, as well as chronic hyperarousal. The definition of hyperarousal is relatively inclusive, with insomnia, hyperactivity, distractibility, racing thoughts, or flight of ideas being acceptable. Furthermore, the child must exhibit abnormal, negative mood between episodes of rage that are chronic and impair functioning in at least two settings. The primary initiative for this diagnosis was to decrease the perceived overdiagnosis of bipolar disorder, though the inclusion of SMD in the DSM and use in the community have been controversial.

This new category generated enough attention in the research community that the DSM-V working group developed a new diagnosis called disruptive mood dysregulation disorder (DMDD) in 2013 that is a modified version of SMD. It differs from SMD in that it does not require chronic hyperarousal symptoms and has fewer exclusionary criteria overall. There are concerns about this diagnosis, however, including limited research as of yet to support the DMDD diagnosis, as the majority of studies pertain to SMD (Axelson et al., 2012). Studies have compared youth with bipolar disorder to those with SMD (Leibenluft, 2011), including a recent review evaluating the similarities and differences between SMD and BP-NOS (Towbin, Axelson, Leibenluft, & Birmaher, 2013). Results show that there are differences in terms of affective response and cognitive flexibility (Dickstein et al., 2007; Guyer et al., 2007), functional neuroimaging (Adleman et al., 2011, 2012), and family history (Brotman et al., 2007). Regarding the DMDD diagnosis, results from the Longitudinal Assessment of Manic Symptoms (LAMS) study (Findling et al., 2010) identified multiple limitations including (1) difficulty differentiating conduct disorder from oppositional defiant disorder, (2) limited diagnostic stability, and (3) its lack of association with current, future onset, or parental history of mood disorders (Axelson, 2013). Overall, initial concerns remain about the diagnostic utility of DMDD and its ability to avoid overdiagnosis of bipolar disorder in children, though more research needs to be performed.

Disruptive Mood Dysregulation Disorder (DMDD)

In order to increase diagnostic latitude and better distinguish affective and behavioral disorders of childhood, the disruptive mood dysregulation disorder (DMDD) diagnosis was created for DSM-5. As previously mentioned, this new diagnosis includes those children who have extreme, recurrent temper tantrums that are out of proportion to environmental stressors and inconsistent with developmental stage. A growing body of research demonstrates that DMDD is distinct from childhood bipolar disorder, which may catalyze additional research into the causes, symptomatology, and treatment of childhood bipolar disorder to help differentiate it from other comorbid illnesses (Leibenluft, 2011; Renk et al., 2014).

Review of Research on the Course of Childhood Bipolar Disorder

Over the past decade, there has been an explosion of research on childhood bipolar disorder (Fristad & Algorta, 2013). The largest increase in number of publications has occurred during this recent time interval, as researchers search for diagnostic

clarity (Fristad & Algorta, 2013). A large number of review papers have been published that seek to consolidate information for the scientific community, with original research papers investigating brain structure, brain function, and pharmacology close behind (Fristad & Algorta, 2013). Multiple longitudinal, DSM-based studies have been conducted, with the most influential being the:

1. "Boston" study (Wozniak et al., 2011)
2. Longitudinal Assessment of Manic Symptoms (LAMS) study (Findling et al., 2010)
3. Early Developmental Stages of Psychopathology (EDSP): Bavarian Catchment study (Beesdo et al., 2009)
4. Course of Bipolar Youth (COBY) study (Birmaher et al., 2006)
5. Assessing Bipolar Disorder: A Community-Academic Blend (ABACAB) study (Youngstrom et al., 2005)
6. "Stanley" study (Findling, Youngstrom et al., 2005)
7. Prepubertal and Early Adolescent Bipolar Disorder (PEA-BD) study (Geller et al., 1998)
8. Oregon Adolescent Depression Project (OADP) (Lewinsohn, Klein, & Seeley, 1995)

These studies are described in detail below.

Influential Longitudinal Studies

Boston Study

The goal of this study was to investigate the longitudinal course of pediatric bipolar I disorder in youth transitioning from childhood to adolescence. Wozniak and colleagues designed a 4-year prospective study of 78 children who were followed into adolescent years, finding that the majority of children continued to experience symptoms of bipolar I disorder into their mid- and late adolescent years. The persistence of symptoms caused great distress in this cohort and was associated with high levels of morbidity and disability. Of note, persistence of subsyndromal forms of bipolar disorder was also associated with dysfunction and morbidity (Wozniak et al., 2011).

Longitudinal Assessment of Manic Symptoms (LAMS) Study

Findling and colleagues designed the Longitudinal Assessment of Manic Symptoms (LAMS) study in order to examine differences in psychiatric symptomatology, diagnoses, demographics, functioning, and psychotropic medication exposure in youth with elevated symptoms of mania (ESM+) compared to children without ESM (ESM-). Guardians of new outpatients 6–12 years of age were asked to complete the Parent General Behavior Inventory-10-Item Mania Scale (PGBI-10 M), and patients with scores ≥12 on the PGBI-10 M (ESM+) and a matched sample of screen negatives (ESM−) were invited to participate. The demographic information, symptom, and diagnostic prevalence, as well as medication exposure for the longitudinal cohort, are discussed in the initial 2010 Findling et al. paper (Findling et al., 2010) although multiple articles on this cohort have been subsequently published (Adleman et al., 2012; Findling et al., 2012). Ten university-affiliated mental health centers participated in the study, with 707 youth participants who were split into either the ESM+ or ESM- groups. Interesting, although ESM+ was associated with higher rates of bipolar spectrum disorders at baseline than ESM-, 75% of ESM+ youth did not meet strict criteria for bipolar spectrum disorders using DSM-IV criteria. This suggests that longitudinal assessment is critical to investigate which factors are associated with advancement to bipolar spectrum disorders in youth with manic symptoms (Findling et al., 2010).

Early Developmental Stages of Psychopathology (EDSP): Bavarian Catchment Study

This pivotal study investigated the incidence pattern of mood episodes in adolescents and young adults living in community settings and the conversion rate from unipolar to bipolar disorder. Beesdo and colleagues conducted a prospective longitudinal community study in order to address three questions: (1) the cumulative incidence of mood episodes and disorders in youth and young adults up to 30 years of age, (2) determine the risk for new-onset depression in individuals with

prior history of hypomanic/manic episodes as well as the risk for new-onset hypomanic/manic episodes in individuals with prior history of depression, and (3) establish the clinical and treatment characteristics of these participants. Overall, 3021 community subjects were enrolled in the study, making this one of the largest studies on this population to date. Results showed that unipolar and bipolar mood disorders are more frequent than previously estimated in adolescence and young adulthood. In those with initial unipolar depression, conversion to bipolar disorder was limited. In those with initial unipolar hypomania/mania, conversion to bipolar disorder was much higher (i.e., 49.6% of the initial unipolar mania cases subsequently developed major depression and 75.6% major or minor depression). Interestingly, bipolar cases did not significantly differ in presenting manic symptoms from unipolar mania cases (Beesdo et al., 2009).

Course of Bipolar Youth "COBY" Study

Birmaher and colleagues conducted a prospective study to investigate the longitudinal course of bipolar spectrum disorders in children and adolescents. Participants were obtained from outpatient and inpatient units at three university centers and included 263 children and adolescents with bipolar spectrum disorders (either bipolar I disorder, bipolar II disorder, or bipolar disorder not otherwise specified (NOS)). The study evaluated rates of recovery and recurrence, weeks with syndromal or subsyndromal mood symptoms, changes in symptoms and polarity, as well as outcome predictors. Results show that children with bipolar spectrum disorders demonstrated a continuum of symptom severity from subsyndromal to full syndromal, with frequent fluctuations in mood. Of note, poorer outcomes and more frequent mood fluctuations were associated with earlier onset, longer duration of mood symptoms, low socioeconomic status, and psychosis. Secondary analysis performed by researchers comparing children and adults with bipolar I disorder showed that children spent significantly more time symptomatic and had more mixed/cycling episodes, mood symptom alterations, and polarity switches than adults. Ultimately, results of this pivotal study provide preliminary support for the bipolar disorder NOS diagnostic category (Birmaher et al., 2006).

Assessing Bipolar Disorder: A Community-Academic Blend Study "ABACAB"

This study by Youngstrom and colleagues was designed to compare the efficiency of six mania measures including the Parent Mood Disorder Questionnaire (P-MDQ), the Adolescent self-report MDQ, the 10-item short form of the Parent General Behavior Inventory (PGBI-SF10), the 28-item Adolescent General Behavior Inventory (AGBI), the Parent-Young Mania Rating Scale (P-YMRS), and the adolescent YMRS in a diverse patient population. Participants included 262 children and adolescents presenting to either an academic institution or community mental health center. Overall, results replicate previous findings that the Parent General Behavior Inventory is the most efficient measure to discriminate bipolar from non-bipolar cases in children and adolescents. This measure is followed by, in order of efficiency, the P-MDQ and the P-YMRS. These three measures appear to be robust in a diverse community setting, while adolescent self-report measures are significantly less efficient at detecting rates of bipolar spectrum disorder (Youngstrom et al., 2005).

Stanley Study

The Findling research group has made significant contributions to the field of childhood bipolar disorder over the past decade. One of their first influential studies, coined the "Stanley" study, sets out to address three issues: (1) describe the phenomenology of children diagnosed with subsyndromal bipolar disorders, (2) describe the phenomenology of children of parents suffering from bipolar disorder who are currently experiencing subsyndromal symptoms of bipolar disorder (i.e., cyclotaxia), and (3) elucidate symptoms useful for identifying children with cyclotaxia. This study included 400 outpatients ranging in age from 5 to 17 years old. Each participant received a diagnostic assessment and dimensional questionnaires to assess mood symptomatology

and psychosocial functioning so that they could be assigned to one of three groups: a syndromal bipolar disorder group, a subsyndromal bipolar group, or a non-bipolar group. Furthermore, based on parental diagnosis, participants were assigned to either a high-risk or a low-risk genetic group. Results demonstrate that children of parents with bipolar spectrum disorder with subsyndromal symptoms demonstrate elevated mood symptoms and psychosocial dysfunction at a greater frequency than youth without this genetic diathesis. In this cohort of children suffering from cyclotaxia, the critical distinguishing characteristics are elevated mood with irritability and rapid mood fluctuations. Prospective follow-up studies are needed for this special population (i.e., offspring of parents with bipolar spectrum disorders), as they are vulnerable to developing bipolar spectrum disorders in the future (Findling, McNamara et al., 2005).

Prepubertal and Early Adolescent Bipolar Disorder "PEA-BD" Study

This study was timely in that it aimed to differentiate prepubertal and early adolescent bipolar spectrum disorders from attention-deficit hyperactivity disorder (ADHD). This was a longitudinal study of the first 60 youth cases diagnosed with bipolarity and the first 60 children diagnosed with ADHD from 270 consecutively ascertained subjects who were each administered comprehensive assessments consisting of the Washington University at St. Louis version of the K-SADS. This assessment tool was developed by the Geller Group at Washington University at St. Louis specifically for use in *prepubertal* evaluations of mania, analogous to those for *prepubertal* MDD, as the traditional K-SADS is not prepubertal specific. Of note, mothers and patients were administered with these interviews blindly by trained professionals. Results found that prepubertal and early adolescent BP cases could be differentiated from ADHD cases by mania-specific criteria (i.e., elated mood, grandiosity, hypersexuality, decreased need for sleep and racing thoughts, but not increased energy and distractibility) and commonly present with ultrarapid or ultradian cycling (Geller et al., 1998).

Oregon Adolescent Depression Project "OADP"

Lewinsohn and colleagues conducted a longitudinal study of the community, examining the prevalence, clinical characteristics, and mental health treatment services utilization of adolescents with bipolar disorder. Structured diagnostic interviews were administered to 1709 adolescents age 14–18 years old, making this one of the largest studies to date. Results identified the lifetime prevalence of bipolar disorders as 1%, with an additional 5.7% of the sample (labeled the "core positive" group) reporting at least one distinct period of abnormally and persistently elevated, expansive, or irritable mood that did not meet criteria for bipolar disorder. Compared with adolescents with a history of major depression and a nonpsychiatric control group, the bipolar and core positive symptom groups exhibited significant functional impairment and high rates of comorbidity, suicide attempts, and utilization of mental health services. This pioneering study demonstrated the clinical significance of subthreshold cases of bipolar disorder in the adolescent population, illustrating the need for appropriate public health resources (Lewinsohn et al., 1995).

A major shift in the research focus on neuropsychiatric disorders in the United States resulted from a National Institutes of Mental Health (NIMH) initiative, as outlined in the 2012 NIMH Strategic Plan that outlines new Research Domain Criteria (RDoC) (Cuthbert, 2014). This document was developed to define new constructs for integrative research purposes, placing emphasis on classifying mental disorders based on dimensions of behavior and neurobiology (Morris & Cuthbert, 2012). The RDoC project ultimately favors an integrative approach to research, including research domains divided into specific constructs that are examined through evaluation of genes, molecules, cells, circuits, physiology, behavior, and self-reports. RDoC research begins with basic mechanisms and investigates dysfunctions in these systems in order to understand symptom sets that span multiple disorders. Through this approach to research, it is hoped that more effective, high-yield treatments will be found (Craske, 2012).

The NIMH intends to alter the focus from DSM diagnostic categories to more dimensional approaches, ultimately accelerating treatment development. This has important implications for our understanding of bipolar disorder in childhood because it may lead to increased understanding of treatment predictors and moderators, which has the potential to facilitate more personalized interventions to monitor treatment response.

Neuropsychology

Literature on adult patients with bipolar disorder consistently indicates neurocognitive deficits in the domains of attention, executive functioning, and verbal memory (Bora, Yucel, & Pantelis, 2009; Martinez-Aran et al., 2004; Thompson et al., 2005), with these impairments persisting at least to some degree in a euthymic state. Overall, evidence suggests that children and adolescents suffering from bipolar disorder present with similar neurocognitive impairments as adults with BPD (Sadock & Sadock, 2007). In particular, youth with bipolar disorder were found to have moderate to severe impairments in the domains of working memory, verbal memory, executive function, processing speed, and sustained attention, compared to healthy controls (Horn, Roessner, & Holtmann, 2011). Additionally, adults with bipolar disorder demonstrate impaired perception of facial expressions of emotion (Benito et al., 2013; David, Soeiro-de-Souza, Moreno, & Bio, 2014). As a result, this area of research was explored in pediatric populations; multiple studies have now demonstrated that children with bipolar disorder also have significant deficits in emotion recognition compared with children who do not carry the diagnosis. Specifically, studies indicate deficits in anger perception in pediatric bipolar disorder patients (Guyer et al., 2007; Perlman et al., 2013; Rich et al., 2006), which is interesting given that anger is an emotion often present in those suffering from this disorder. Recognition and management of these deficits is critical, as it can impact interpersonal skills, academic performance, and development of self-esteem in youth.

Structure and Functional Neuroimaging Findings

Functional and structural neuroimaging studies have shown that there are several brain regions that are implicated in the development and maintenance of bipolar symptoms in children and adolescents. Similar to adult studies, for which more empirical data from neuroimaging studies exist, research on children and adolescents with familial bipolar disorder suggests a role for disruption of subcortical structures. Specifically, structural neuroimaging studies have found children with familial bipolar disorder to have smaller amygdala volumes relative to control children (Blumberg et al., 2003; Chang et al., 2005; DelBello, Zimmerman, Mills, Getz, & Strakowski, 2004). Recent studies also found that children with bipolar disorder exhibit differences in the volume of the hippocampus, caudate, and/or thalamus relative to healthy controls (Chang et al., 2005). One study found that children and adolescents who had experienced at least one manic or hypomanic episode had reduced gray matter volume in the left dorsolateral prefrontal cortex, the left accumbens, and the left amygdala relative to healthy controls (Dickstein et al., 2005), while other studies found that adolescents with bipolar disorder demonstrated decreased volume in the left superior temporal gyrus (Chen et al., 2004; Pfeifer, Welge, Strakowski, Adler, & DelBello, 2008). Notably, medications also appear to have significant effects on brain structure in both children and adults, for example, Chang and colleagues found a relationship between exposure to valproate and lithium and increased gray matter volume in the amygdala (Chang et al., 2005).

Expanding from brain structure to brain function, functional neuroimaging studies suggest that children and adolescents with bipolar disorder may have abnormalities in prefrontal-subcortical circuits. Chang's research group administered a visuospatial working memory task to children and adolescents with familial bipolar disorder vs. typically developing controls, finding that those with bipolar disorder had greater activation in the bilateral anterior

cingulate cortex, left putamen, left thalamus, left dorsolateral prefrontal cortex, and right inferior frontal gyrus while control subjects had greater activation in their cerebellar vermis (Chang et al., 2004). Additionally, in this study, during an affective task involving the visualization of positively valenced pictures in particular, children with bipolar disorder had increased activation in frontal and striatal regions (caudate/thalamus), as well as frontal and anterior cingulate regions. Given the role of the anterior cingulate cortex and the dorsolateral prefrontal cortex in normal mood regulation and sustained attention (George et al., 1995), these patterns suggest dysregulation of these circuits in children with familial bipolar illness.

Moreover, there is increasing evidence of reduced functional connectivity between prefrontal regions and limbic regions within affective and working memory networks in pediatric bipolar disorder. Specifically, while undergoing fMRI imaging during an affective working memory task involving labeling of neutral and angry facial expressions, pediatric bipolar disorder patients demonstrated decreased functional connectivity relative to healthy controls in brain regions involved in emotion processing (right amygdala) and in emotion regulation (right ventrolateral prefrontal cortex), while connectivity was increased in brain regions involved in the assessment of emotion (bilateral medial prefrontal cortex) (Ladouceur et al., 2011).With respect to affective working memory networks (i.e., amygdala, temporal lobes, and dorsolateral and ventrolateral prefrontal regions (Strakowski, 2015)), those with pediatric bipolar disorder demonstrate greater connectivity compared to healthy controls in the left dorsolateral prefrontal cortex, caudate, and right ventrolateral prefrontal cortex, with reduced connectivity between emotion-processing regions (right amygdala and bilateral temporal regions). Overall, these findings suggest disrupted neural connectivity during tasks involving both affective and frontal executive components (Ladouceur et al., 2011; Passarotti, Ellis, Wegbreit, Stevens, & Pavuluri, 2012). Further investigation of brain circuitry, both at rest and during participation in tasks of cognition and emotion, must be conducted in this population in order to make concrete conclusions (Faraone et al., 1997).

Structural Neuroanatomy

The structural neuroanatomic substrates of childhood bipolar disorder are an area of increased investigation. While brain volume reaches approximately 90% of its adult size by 6 years of age, MRI studies show that myelination and overall white matter development persist well into the fifth decade of life (Walhovd, Johansen-Berg, & Karadottir, 2014). As mentioned earlier, a growing body of research has focused on abnormal development of subcortical structures and prefrontal-subcortical circuits in children with bipolar disorder. Specifically, studies have demonstrated smaller amygdala volumes; alterations in hippocampal, caudate, and thalamic volumes; and reduced gray matter volume in the left dorsolateral prefrontal cortex, left accumbens, left amygdala, and left superior temporal gyrus in children with bipolar disorder (Dickstein et al., 2005).

Results of these neuroimaging studies are far from diagnostic at this stage, though certainly warrant increased investigation as they may help identify vulnerable populations who will benefit from close follow-up and intervention (Sadock & Sadock, 2007).

Etiology

Historically, bipolar disorder was thought to rarely occur in youth. Recent evidence challenges this notion, as there has been a significant increase in children and adolescents diagnosed with bipolar disorder (Blader & Carlson, 2007). A recent meta-analysis including 12 studies from around the world from 1985 to 2007 demonstrated that the overall prevalence of bipolar disorder in children and adolescents was 1.8% (Van Meter, Moreira, & Youngstrom, 2011). Yet, the most valid diagnosis for youth with mood lability, irritability out of proportion to stressor, or rapid mood cycling remains controversial.

Retrospective studies have found that among the 20–30% of adults with bipolar disorder who exhibit "mixed mania (i.e., mania with concurrent depressive symptoms)," higher rates of childhood onset are present. These individuals are found to have a chronic course, absence of discrete episodes, higher risk of suicidal behavior, and a poorer response to treatment. These presenting symptoms are similar to the clinical presentation of prepubertal children who are described as having atypical bipolar disorders (Sadock & Sadock, 2007).

Genetics

Family studies demonstrate that children of a parent with bipolar I disorder have a 25% chance of developing a mood disorder, while children of two parents with bipolar I disorder have a 50 to 75% risk of developing a mood disorder. The high rates of comorbid ADHD among children with bipolar disorder have caused researchers to investigate the co-transmission of these disorders in families. Children of parents with bipolar disorder have been found to have higher rates of ADHD compared with controls. In first-degree relatives of children with bipolar disorder, ADHD occurs with the same frequency as in first-degree relatives of children with ADHD only. However, the combination of ADHD and bipolar disorder was not found as frequently in relatives of children with ADHD only, compared with first-degree relatives of children with comorbid illness. Altogether, these results suggest that pediatric bipolar disorder may represent an early-onset form of bipolar disorder with a strong genetic diathesis, including psychiatric comorbidities such as ADHD (Sadock & Sadock, 2007).

Course and Developmental Challenges

Bipolar spectrum disorders in the adult and pediatric population are enduring, with symptoms that are typically chronic, progressive, and recurring (Wozniak et al., 2011). Although bipolar spectrum disorders are thought to be episodic in nature, they can present with or without inter-episodic recovery. In a multicenter study with a duration of 4 years, 40% of children and adolescents with bipolar spectrum disorders experienced syndromal or subsyndromal symptoms during 75% of the four-year follow-up period (Birmaher et al., 2009). Mixed symptoms (46%) and symptoms of depression (33.8%) were more prevalent than symptoms of mania (21%), and approximately 40% of children diagnosed with bipolar disorder not otherwise specified converted to bipolar I or II disorders. While 80% responded to treatment and recovered from their initial episode, almost two-thirds experienced recurrent episodes with the majority being depressive episodes. In fact, approximately 40% of youth with bipolar spectrum disorders remain chronically ill with impaired functioning or with a slowly improving course (Birmaher et al., 2014). As children with bipolar disorder advance through puberty, symptoms of aggression, irritability, and excessive motor activity decrease (Demeter et al., 2013; Safer, Magno Zito, & Safer, 2012), while the frequency of depressive symptoms typically increases (Demeter et al., 2013). With respect to gender differences, although the overall clinical presentation is not dramatically different between female and male children with bipolar disorder, females tend to have a shorter duration of mania, increased frequency of depressive episodes, and increased vulnerability to comorbid panic disorder (Wozniak et al., 2013). Males, on the other hand, have an increased frequency of comorbid disruptive behavioral disorders such as ADHD and an increased frequency of hypersexuality after puberty (Geller, Zimerman et al., 2000).

The World Health Organization recently listed bipolar spectrum disorders as the fourth leading cause of disability among adolescents in the world (Gore et al., 2011). This supports previous studies concluding that children and adolescents with bipolar spectrum disorders have a lower quality of life than children with other chronic medical conditions and mental illnesses (Freeman et al., 2009). As discussed

previously, these children tend to struggle in the academic setting, which is compounded by extreme difficulty with social interactions (Geller, Bolhofner et al., 2000). Data suggest that childhood abuse and neglect are risk factors associated with a worsening clinical course of bipolar disorder in this already vulnerable population (Daruy-Filho, Brietzke, Lafer, & Grassi-Oliveira, 2011). Alarmingly, youth with bipolar spectrum disorder are more likely to engage in self-injurious or suicidal behavior (Goldstein, 2009), making proper diagnosis and timely treatment critical.

Treatment

Prior to administration of psychotropic medications, treatment of bipolar disorder in children and adolescents involves consideration of multiple variables. Clinicians must account for the effects of normal development, age-related clinical presentations, comorbid psychiatric disorders, and age-specific differences in the efficacy and tolerability of psychiatric medications (Ketter, 2010). When working with this special population, clinicians must consider family environments, parental practices, and educational settings in order to maximize positive outcomes. Successful interventions with the best long-term outcomes often involve a multidisciplinary team consisting of a child and adolescent psychiatrist, psychologist, school counselor, parents, and educational specialists. Medications are certainly paramount in stabilizing children and adolescents acutely, while maintenance often involves multiple forms of treatment. As discussed previously, in contrast to the usual argument for monotherapy in pediatric populations, combination treatments are often found to be necessary (Kowatch, Sethuraman, Hume, Kromelis, & Weinberg, 2003). Research has advanced to studying combination algorithms involving either multiple mood stabilizers (Kowatch et al., 2003) or mood stabilizers augmented with atypical antipsychotic medication (i.e., risperidone, olanzapine, quetiapine, ziprasidone, and aripiprazole) (Pavuluri et al., 2006).

Mania

Medications with FDA approval for acute treatment of childhood bipolar disorder/mania are lithium (age 12 years and older) and the atypical antipsychotics aripiprazole, risperidone, quetiapine (all for ages 10 years and older), and olanzapine (age 13 years and older). Considering that youths with bipolar disorder commonly experience more weight gain with antipsychotic medications than adults, olanzapine should be reserved for those who have failed other medication trials. Compared with adults, children and adolescents with mania have greater response to the atypical antipsychotics and experience lower effect sizes for treatment response with the mood stabilizers divalproex and lithium. In an open-label, 12-month study of preschool-onset bipolar disorder conducted by Pavuluri and colleagues, risperidone worked faster and more effectively than divalproex in reducing symptoms of mania (Pavuluri et al., 2010). For maintenance treatment of mania, the goal is persistent mood stabilization and preventing recurrence of episodes. Important components in this long-term phase of treatment involve assessment of residual symptoms, treatment adherence, and side effects (Krieger & Stringaris, 2013). A long-term, randomized, 30-week trial in youth with bipolar disorder showed that aripiprazole was superior to placebo in improving mania symptoms, though more research needs to be done utilizing head-to-head, randomized double-blind comparison studies (Findling et al., 2012).

Depression

To date, there is no FDA-approved medication for bipolar depression in children and adolescents. Most of the data on this topic has been extrapolated from research on the adult population, with more anecdotal results than evidence-based-driven studies. Quetiapine has been effective in treating bipolar depression in adults and has been used in the child and adolescent population with reasonable efficacy (Krieger & Stringaris, 2013). Antidepressant monotherapy,

however, is generally not used in bipolar depression until treatment with a mood stabilizer is established, for fear of precipitating mania (Dulcan & Lake, 2012). Both cognitive-behavioral therapy and family-focused therapy have been shown to work well in this population, whether used alone or in combination with pharmacologic agents.

Pharmacology Research

Pharmacologic studies have increased as scientists search for effective treatments for children with bipolar disorder. Results have led to algorithms supporting the use of atypical antipsychotics ahead of mood stabilizers, though the side effect profile of all psychotropic agents must be carefully considered (Correll, Sheridan, & DelBello, 2010; Goldstein, Sassi, & Diler, 2012). Of note, olanzapine is approved for children age 13 and older, though is usually used after multiple failed trials of other antipsychotic medications due to predisposition to weight gain (Dulcan & Lake, 2012). Research has focused on targeting specific agents to be used alone and in combination, in hopes of providing clinicians with clear guidelines. In a controlled, randomized study of 279 children and adolescents with bipolar I disorder, Geller and colleagues found a significantly greater response rate (as measured by the Clinical Global Impressions for Bipolar Illness Improvement-Mania Scale) and fewer side effects (as assessed by the Modified Side Effects Form for Children and Adolescents) to the antipsychotic risperidone than to mood stabilizers lithium and divalproex sodium (Geller et al., 2012). Clinicians must consider the risk of metabolic syndrome in youth, however, who are exposed to antipsychotic medications for multiple years (Geller et al., 2012). As such, research has advanced from investigating monotherapy to combination algorithms involving either multiple mood stabilizers (Kowatch et al., 2003) or mood stabilizers augmented with atypical antipsychotic medication (i.e., risperidone, olanzapine, quetiapine, ziprasidone, and aripiprazole) (Pavuluri et al., 2006). Findling et al. studied response to treatment with a combination of lithium and divalproex sodium in children and adolescents meeting criteria for bipolar I disorder or bipolar II disorder (Findling et al., 2003). Once stabilized on these medications, some children were able to remain symptom-free, while others experienced relapse or adverse events (Findling, McNamara et al., 2005). In those who relapsed or experienced intolerable side effects, atypical antipsychotics were systematically introduced that were specific to each child with respect to side effect profile (Kowatch, et al., 2005; Pavuluri et al., 2004). Results using this algorithm approach reduced symptoms of bipolar disorder (Pavuluri et al., 2004), though more targeted research is necessary.

The majority of pharmacologic research on bipolar disorder in children has investigated individuals 5 and older (Vitiello, 2001; Zito et al., 2000). In the few studies that were conducted with children of preschool age, several of the commonly used medications that were efficacious in older children also appeared to be effective in this cohort. Specifically, through the use of the Young Mania Rating Scale (YMRS) as the primary outcome measure with response defined as a $\geq$50% decrease from baseline, evidence was found for use of atypical antipsychotics (risperidone, olanzapine, quetiapine), as well as mood stabilizers (valproate, divalproex sodium, and carbamazepine) in younger children (Biederman et al., 2005; Joshi et al., 2012; Mota-Castillo et al., 2001; Scheffer & Niskala Apps, 2004; Tuzun, Zoroglu, & Savas, 2002). Lithium, on the other hand, may not be as effective as the other medications, though more controlled studies are needed (Pavuluri et al., 2006). Most importantly, side effect profile, ease of administration and dosing, as well as cost must all be considered when selecting medications.

Psychosocial Treatment

Family-focused treatment (FFT) has been shown to enhance maintenance of stable mental health in children and adolescents with bipolar disorder when used in combination with optimized

psychopharmacology (Miklowitz et al., 2008). FFT is a psychoeducational intervention created to reduce familial stress, conflict, and affective arousal by optimizing communication and problem-solving between patient and caregivers (Krieger & Stringaris, 2013). The goals for this treatment protocol focus on increasing the understanding of mood symptoms, improving medication compliance, enhancing communication skills, and reducing impairments in social functioning (Renk et al., 2014). Recent research found evidence that this treatment significantly reduced bipolar disorder symptoms in adolescents (Miklowitz, Biuckians, & Richards, 2006). Furthermore, the child- and family-focused cognitive-behavioral therapy (CFF-CBT) protocol was developed as an extension of FFT for children ranging in age from 8 to 12 with their families. The treatment protocol incorporated cognitive-behavioral techniques, interpersonal skills, and psychoeducation with patients, parents, parent-patient dyads, siblings, and school counselors, thus targeting those most directly affected by the illness. Research demonstrated symptom reduction when this treatment was provided both individually (Pavuluri et al., 2004) and in a group (West et al., 2009) setting. Even more impressive, these gains were sustained when a maintenance phase was added to the treatment, which included booster sessions and suggestions for medication management (West, Henry, & Pavuluri, 2007). Interestingly, in child cohorts with bipolar depression, FFT has shown promising results. In a randomized trial comparing FFT with enhanced care in addition to pharmacotherapy in adolescents, the FFT group recovered faster from depressive symptoms, spent less time in depressive episodes, and had an improved outcome for 2 years (Miklowitz et al., 2006).

Preliminary results on cognitive-behavioral therapy (CBT) for bipolar depression demonstrate a significant decrease in depressive symptoms (Krieger & Stringaris, 2013). Given the large pool of data for effectiveness of CBT in unipolar depression, clinicians have applied this structured therapeutic approach to children and adolescents with bipolar depression. One example is the "Think Effectively About Mood Swings (TEAMS)" program (Searson, Mansell, Lowens, & Tai, 2012), which is based on an integrative cognitive model. This model suggests that multiple and extreme appraisals of internal states and the impact these changes have on behavior actually maintain and exacerbate bipolar symptoms. By addressing these extreme positive and negative appraisals of internal states through CBT, bipolar symptoms were improved (Searson et al., 2012). Although this program was investigated in a small adult sample, such programs can be adapted to the needs of children at their appropriate developmental level. The use of CBT in combination with pharmacotherapy or alone, however, should carefully consider level of impairment and cognitive resources of the patient (DeFilippis & Wagner, 2013).

Prognosis

There are multiple factors that affect prognosis of childhood-onset bipolar disorder. Studies have consistently demonstrated that the longer the illness goes undiagnosed and consequently untreated, the poorer the prognosis (Leverich et al., 2007). Given the diagnostic controversy deriving from the lack of discrete episodes, comorbidity with other childhood disorders, and hesitancy to initiate mood stabilizers and/or antipsychotic medications in childhood (Leverich et al., 2007), the psychiatric community recognizes the need to standardize diagnosis. Moreover, clinicians recognize the need to advance childhood-onset bipolar disorder from a diagnosis of exclusion to one that is carefully considered in all children and adolescents presenting with concerning symptoms.

Factors associated with poorer outcomes include a DSM-V diagnosis of bipolar disorder specified other or unspecified other, early onset, low socioeconomic status, and family history of mood disorders (Singh et al., 2014). Specifically, Leverich et al. (2007) and Perlis et al. (2004) demonstrated that earlier age of onset was associated with a more difficult and complicated course of illness, including faster cycling, more days depressed, greater lifetime manic and depressive

episodes, increased risk of substance abuse and other comorbidities, and greater lifetime risk of suicide attempts. Individuals with lower socioeconomic status and therefore decreased access to mental health resources are naturally at a disadvantage, often lost in an already complicated system. Pediatricians play an integral role in improving rates of diagnosis and treatment, as they are the gatekeepers of childhood medical care and may be able to recognize cardinal symptoms of childhood-onset bipolar disorder. If this diagnosis is in the differential, appropriate referrals to child and adolescent psychiatrists are warranted. The earlier bipolar disorder is diagnosed and appropriately treated, the better the individual outcome.

Outcome

Similar to prognostic studies, outcome studies are particularly interested in understanding the contribution of bipolar disorder symptoms to adult functioning. As stated above, multiple studies have demonstrated that early onset in bipolar disorder has been associated with greater familial risk and unfavorable clinical outcomes. Symptoms that challenge youth beyond the difficulties presented during puberty and normal development threaten maturation and identity development (Baldessarini et al., 2012). A study including youth from six different countries who were diagnosed with bipolar disorder found significant impairments in adult functioning as measured through higher rates of unemployment, not living independently, and being unmarried without children (Baldessarini et al., 2012). In combination with medication management and access to appropriate therapy (i.e., family based versus CBT), youth require interventions that limit maturational-functional impairments as they develop.

Another study conducted by Birmaher and colleagues reported that while most children with early-onset mania recovered within 2 years, approximately 80% experienced recurrences over the next 2–5 years (Birmaher et al., 2009). Over a follow-up period of 4 years, approximately 30% remained euthymic, 40% had continuing substantial symptoms, and 20% remained seriously ill. Alarmingly, Birmaher's data indicate that those with childhood-onset bipolar illness remained symptomatic during 60% of the follow-up period.

Approximately 4 years later, Birmaher published results demonstrating a more hopeful outlook (Birmaher et al., 2014). Specifically, he studied long-term mood trajectories in youth with bipolar disorder with an emphasis on factors relevant to persistent euthymia during the follow-up period. After 8 years of follow-up, four different mood trajectories were identified in youth: (1) "predominantly euthymic," being euthymic the majority (>80%) of the follow-up period (N = 88); (2) "moderately euthymic," being euthymic about half of the follow-up period (N = 127); (3) "ill with improving course," being euthymic less than half of the follow-up time (N = 70); and (4) "predominantly ill," being euthymic for only a minority (~11%) of the follow-up period (N = 82). History of poor functioning was similar across all four classes, and all four classes had high rates of comorbid psychopathology, pharmacologic interventions, history of abuse, poor family functioning, and family history. Six variables that translated into a better longitudinal course in youth were later age at onset of mood symptoms, lower severity of depression, lower frequency of manic or hypomanic symptoms, fewer subsyndromal episodes, fewer suicide attempts, less history of sexual abuse, and less family history of bipolar disorder or substance use disorders. This study reveals a more optimistic outlook on a chronic illness that was once thought to be completely debilitating. This study demonstrates that approximately 43% of the sample was euthymic most of the time, though taking into consideration that over 50% of those in the predominantly euthymic group continued to experience active, bipolar symptoms. All in all, this study and the current literature support the notion that childhood-onset bipolar disorder, while challenging and potentially resulting in significant functional disability, is a mental illness that is treatable. Using a robust diagnostic arsenal consisting of the DSM-V and thorough assessment over a period of time, clinicians can feel confident

about their differential, which will hopefully translate into earlier diagnosis and treatment for this vulnerable population.

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Depressive Disorders

7

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General Overview

Much of the history of childhood depression as a diagnostic construct is characterized by disagreement. For many decades, theorists and researchers argued whether children were capable of experiencing depression. Researchers observed comparatively lower rates of depression in children when compared to adults, such that many believed that depression did not exist for children. As the discussion evolved, attention was turned toward whether the manifestation of depression differed in children when compared to adults.

The first and second editions of the *Diagnostic and Statistical Manual of Mental Disorders* were heavily influenced by the dominant psychoanalytic theories of the time (Mayes & Horowitz, 2005). Within this school of thought, depression was understood to be the result of feelings of anger and hostility turned inward, toward the self, by the superego (Finch, 1960; Rie, 1966). For older adolescents and adults, the experience of depression was thought to be rooted in the combination of an individual's strict superego and unconscious infantile characteristics (Finch, 1960). Due to their immaturity, these individuals desire more than can be given by their environment, which results in feelings of frustration and resentment. The superego prevents them from outwardly expressing the negative emotions and directs the feelings inward, which manifest as hopelessness, worthlessness, and self-deprecation (Finch, 1960). However, for young children, whose superegos are not yet fully developed or internalized, they are unable, as it was believed, to redirect negative emotions and, therefore, unable to experience depression.

Some psychoanalytic theorists contended that children were unable to experience depression, while others theorized that childhood depression existed, but possessed an intrapsychically disparate structure than adult depression (Rie, 1966). Psychoanalysts who maintained that depressive experiences could occur during childhood observed evidence of depressive symptoms in young children and infants. Spitz (1945) described a condition he termed "anaclitic depression" in infants who were separated from their mother in their first 5 years of life and placed in institutional care. He noted that infants showed a number of behaviors characteristic of depression, including appearing sad and apprehensive, withdrawing from contact with others, and a lack of appetite.

Throughout the 1950s and early 1960s, the lack of agreement over the existence of childhood depression within the field likely contributed to

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childhood depression being given lesser levels of attention in the literature. Although the existence of childhood depression was contested, there was considerable consensus regarding the absence of the familiar symptoms of adult depression in children. As noted above, some suggested that this was because depression was not experienced by children. Others theorized that the disparity was due to developmental differences in the way symptoms manifested that were unaccounted for in the conceptualization of depression (Lesse, 1983). These researchers argued that children would express symptoms in age-appropriate ways that could differ substantially from symptoms in adults; thus, there was an underlying core construct of depression which the field had yet to accurately identify. Based on this argument, many symptoms were theorized to be developmentally appropriate expressions of depression. These symptoms were called "depressive equivalents," and the syndrome was referred to as *masked depression* (Cytryn & McKnew, 1972; Lesse, 1983). Masked depression became the label for a variety of somatic complaints and behavioral disturbances that were posited as being a coping response to depression (Lesse, 1983).

As a depressive construct with an emphasis on somatic symptoms, masked depression described clients who complained of physical discomfort and denied psychopathological difficulties such as depressed mood, but whose etiology was believed to be rooted in depression. For young children in particular, it was theorized that most would not be able to verbalize their depressive experiences, but would act in ways more appropriate to their developmental level. Children and adolescents who were diagnosed with depression also showed high levels of co-occurring behavioral difficulties and other symptoms that were not associated with adult depression, such as enuresis and school refusal. Within the criteria for a diagnosis of a major depressive episode in the *Diagnostic and Statistical Manual for Mental Disorders, 3rd Edition* (DSM-III; American Psychiatric Association [APA], 1980), there was an age-specific note that depressed mood could be inferred by a sad facial expression for children and adolescents. This was added to capture the potentially developmentally limited capacity for children and adolescents to verbalize their depressive symptomatology.

These externally observable symptoms were considered by many within the field to be overlying a core internalized experience of depression. However, no reliable or valid criteria were formed for symptoms of depressive equivalents or for the diagnosis of masked depression (Angold, 1988). Indeed, it seemed inherent to the construct of masked depression that the variety of ways in which depression could indirectly manifest required subjective interpretation and therefore was unable to be conceptualized through objective criteria. Within a short period of time, the boundaries for what could be considered an overlying symptom masking depression soon expanded to encompass nearly all of childhood psychopathology (Angold, 1988). The acceptance of depressive equivalents and the consequent inflation of symptoms that could be representative of a child or adolescent's depression contributed to further difficulties in clarifying and substantiating childhood depression as a valid psychiatric diagnosis. The overwhelming number of potential masked symptoms pushed the boundaries defining depression past the point of utility. As a result, masked depression was rejected as a diagnostic construct and is no longer considered a valid diagnosis (Angold, 1988).

Although the existence of depression within childhood gained increasing research support during this time, many clinicians continued not to diagnose depression for children, even when significant depressive symptoms were present (Angold, 1988; Friedman et al., 1982). Pervasive, yet inaccurate, cultural ideas of the lability of adolescents' mood, in addition to the overdiagnosis of schizophrenia in adolescents presenting depressive symptomatology, contributed to the low rates of depressive diagnoses (Carlson & Strober, 1978).

As acceptance for the experience of childhood depression grew, researchers turned their focus toward parsing out the symptomatology of depression within children. There was considerable dissent regarding the utility of a separate set of diagnostic criteria for children and adolescents. Some researchers found that the established adult criteria were able to identify children and adolescents with depression (Mitchell, McCauley, Burke, & Moss, 1988). However, others had concerns about whether the unmodified criteria of

depression for adults would accurately capture depression in youth populations.

Consensus regarding the existence and symptomatology of childhood depression was not reached until the late 1970s, with the work of Puig-Antich, Blau, Marx, Greenhill, and Chambers (1978), Carlson and Cantwell (1980), and others who demonstrated that many children meet the full diagnostic criteria for major depressive disorder in adults. The results of these studies demonstrated that while the "masked" symptoms of depression in children and adolescents were present, they did not exist in isolation. These youth also exhibited typical depressive symptoms associated with depression in adults. Depressive equivalents were then described to be overt, more easily noticeable concerns, which prevented clinicians from accurately identifying the less obvious, but extant, depressive symptoms.

Major Depressive Disorder (MDD)

The diagnostic criteria for major depressive disorder have remained relatively consistent through the DSM-III to the current DSM-5 (American Psychiatric Association [APA], 2013). There is one set of criteria for both adults and children, based on research findings that children and adolescents with depression can be reliably identified using the same criteria; however, irritable mood was added for children and adolescents as a possible alternative for an essential feature of major depression instead of depressed mood. This feature has been retained in the DSM-5 and allows irritable mood to substitute the presence of depressed mood in children and adolescents. Irritability is more commonly seen in children and adolescents, but most children with depression will exhibit depressed mood either alone (58%) or combined with irritable mood (36%) and rarely will display only irritable mood (6%) (Stringaris, Maughan, Copeland, Costello, & Angold, 2013).

Following DSM-5 criteria, a diagnosis of major depressive disorder (MDD) requires the presence of a major depressive episode. These episodes require that a child or adolescent exhibits one of the two key features: depressed or irritable mood and a loss of interest or pleasure. These symptoms are accompanied by additional depressive symptoms including significant weight loss or weight gain, insomnia or hypersomnia, and feelings of worthlessness, which must be present most days within the same 2-week period. Importantly, the presence of certain exclusionary factors that could cause or contribute to the depressive symptoms must be ruled out. These include the effects of substances, medical conditions, and depression that occurs as part of the typical experience of significant loss.

After assessing for exclusionary conditions, if a youth meets the full criteria for a diagnosis of MDD, the DSM-5 includes dimensions of severity that should be included in the diagnosis ("mild," "moderate," "severe"). These ratings are based on the number of symptoms an individual displays beyond what is needed for the diagnosis, the associated distress of the condition, and the level of impairment that it causes.

Persistent Depressive Disorder (P-DD)

Persistent depressive disorder is a new diagnostic category in the DSM-5 that consolidates two preexisting diagnostic categories: chronic major depression (MDD-chronic) and dysthymic disorder (APA, 2013). While much of the research on childhood depressive disorders has concentrated on major depressive disorder, the distinctions between MDD-chronic and dysthymic disorder have been reexamined.

Dysthymic disorder was added to the DSM nosology in 1980 with the publication of the DSM-III. For children and adolescents, dysthymic disorder, or dysthymia, is a depressive disorder characterized by depressed mood for most of the day, more days than not, lasting for at least 1 year. Although these symptoms persisted over long periods of time, fewer symptoms are required to meet the criteria for the diagnosis, and they are often milder than the symptoms experienced in major depressive disorder. Although earlier versions of the DSM maintained distinct diagnoses for several types of chronic depressive symptoms, research has not found clinically or scientifically meaningful dif-

ferences between dysthymic disorder and MDD-chronic (McCullough et al., 2003). Chronic depression, whether conceptualized as dysthymic disorder or MDD-chronic, is associated with very similar characteristics including poorer response to treatment. Thus, these categories were combined to create the overarching category *persistent depressive disorder*.

A diagnosis of P-DD requires that a child or adolescent exhibit depressed or irritable mood most of the day, for most days, over a period of at least 1 year. During this period of depression, youth must also display two or more depressive symptoms (e.g., insomnia or hypersomnia, low self-esteem, feelings of hopelessness). For some children and adolescents, they may experience "double depression," where they experience a major depressive episode on top of preexisting P-DD. In these situations, these youth are diagnosed with both major depressive disorder and P-DD.

Disruptive Mood Dysregulation Disorder (DMDD)

In addition to the newly named P-DD, DSM-5 introduced a new classification to childhood depressive disorders labeled disruptive mood dysregulation disorder. This disorder captures nonepisodic emotional and behavioral problems for children 6 years of age or older, with an onset that occurred prior to age 10. The key feature of this disorder is severe, chronic irritability. This disorder was introduced to the DSM-5 following concerns surrounding the drastic increase in diagnoses of childhood bipolar disorder. The widespread increase in diagnoses of bipolar disorder was troubling particularly given the frontline treatment provided. Treatment for bipolar disorder often includes the use of antipsychotic medication, the effects of which are not fully understood within children and adolescents, particularly as they relate to impacts on development.

Historically, bipolar disorder has been viewed as a condition that can initially develop during childhood, but that does so rarely. However, within the past 20 years, there has been a dramatic increase in the number of preadolescent children diagnosed with bipolar disorder (Moreno et al., 2007). Between 1993 and 2003, while diagnoses of bipolar disorder in adult populations doubled, individuals under 20 years of age were diagnosed with bipolar disorder 40 times more frequently (Moreno et al., 2007). This increase in diagnoses could be the result of either better detection of bipolar disorder by clinicians among younger populations or the application of a less stringent definition of bipolar disorder within this population (Friedman et al., 1982). The research in this area has produced conflicting observations (Pogge et al., 2001). While the recent and rapid increase of bipolar diagnoses among children and adolescents is a trend that cannot be disputed, there is little consensus regarding the causal and moderating factors.

In 2003, Leibenluft and colleagues proposed the diagnosis *severe mood dysregulation* (SMD) to better capture the symptomatology believed to be adding to the inflation of bipolar diagnoses among children and adolescents. The core set of symptoms included chronic and severe irritability and symptoms of hyperarousal. Importantly, children and adolescents who display persistent irritable mood in addition to symptoms of hyperarousal associated with episodes of mania fall into a gray area of clinical understanding. Many of the symptoms these youth demonstrate appear characteristic of bipolar disorder; however, the pervasive irritable mood patterns exhibited are drastically unlike the defined, episodic quality of elevated or irritable mood that defines a manic episode (Leibenluft, 2011).

Severe mood dysregulation was added to the DSM-5 under the label *disruptive mood dysregulation disorder* (APA, 2013). To meet the criteria for this diagnosis, children and adolescents must display the core feature of chronic, severe, persistent irritability. This irritability is operationalized with two main characteristics. First, youth must exhibit frequent and developmentally inappropriate temper outbursts. Second, between temper outbursts, youth must exhibit irritable or angry mood most of the day, nearly every day. These characteristics must be present before age 10 years and last for at least 1 year.

As a new disorder to the DSM-5, disruptive mood dysregulation disorder is a condition about which relatively little is known. Preliminary information regarding the impact of the inclusion of DMDD in the DSM-5 on the rate of bipolar diag-

noses among youth has provided some evidence that the rates of inaccurate bipolar diagnoses are reduced by using DMDD criteria; however, this is an ongoing area of research (Margulies, Weintraub, Basile, Grover, & Carlson, 2012).

Etiology

Prevalence

Exact rates of childhood depressive disorders are difficult to discern. Studies that focus on preadolescent children with DSM diagnoses are limited, and young children are not typically included in large-scale community surveys (Mash & Barkley, 2003). However, it is estimated that as many as one in 34 children and one in eight adolescents are diagnosed with depression (Merikangas et al., 2010; Perou et al., 2013). Although information on rates of depression within adolescent populations is also limited, a nationally representative epidemiological community survey within the US estimated lifetime prevalence of MDD in 13- to 18-year-olds was 11% (Avenevoli, Swendsen, He, Burstein, & Merikangas, 2015).

Biological Vulnerability

As with adults, the development of depressive disorders in children and adolescents has not been attributed to a single cause. Research has identified multiple factors affecting whether a youth will develop a depressive disorder, such that contemporary models of depressive disorders require an integration of neurobiological, cognitive, interpersonal, and genetic factors. Given the large number of potential factors which influence each other, it is likely that there is not a single cause or pathway to a depressive disorder, but multiple pathways from which a depressive disorder could develop.

Estimates of the heritability of MDD based on twin studies range between 40% and 50% (Bierut et al., 1999; Kendler, Gardner, Neale, & Prescott, 2001). Despite the significant sex differences in the prevalence of depressive disorders after puberty, a recent meta-analysis found small to no differences in genetic etiology, from which the authors concluded that the heritability of depressive disorders is likely not associated with genetic differences among the sexes (Franić, Middeldorp, Dolan, Ligthart, & Boomsma, 2010). However, findings in this area have been inconsistent, with some studies showing none to moderate sex differences and discrepant findings regarding whether males or females appear to be more affected by genetics than the other sex (Lau & Eley, 2013).

MDD has been shown to aggregate within families, where first-order relatives are at an elevated risk for developing depression (Weissman et al., 2005). In addition to higher risk, familial depression is associated with an early age of onset and recurrence into adulthood (Wickramaratne, Warner, Weissman, 2000). Children of parents with depression are three times more likely to develop MDD than children of nondepressed parents (Weissman et al., 2005). In one of the longest longitudinal studies of the familial influence on depression, the authors observed three generations over a period of 30 years. The grandchildren of the original participants who were depressed in the initial sample were at higher risk for developing MDD (Weissman et al., 2005).

Although some studies have noted significant effects when both fathers and mothers are depressed, the relationship between maternal depression and a wide range of childhood psychopathology has been a focus of many decades of research. Children of depressed mothers are more likely to develop internalizing disorders and behavioral issues (Goodman et al., 2011). These children are also more likely to display high levels of negative emotionality and low positive emotionality, both traits associated with higher risk of developing depression (Klein, Durbin, & Shankman, 2009). Though this association has been well supported in research, the specific causes underlying this association are not yet clearly understood. It is likely the result of a number of complex interactions including genetics, neurobiology, and environmental factors.

Despite the extant research on the effects of maternal depression on childhood and adolescent depression, many areas simply require more research. When mothers are depressed, there are

often environmental factors involved, including marital discord, stressful work or economic situations, and negative interactions with children (Hammen, Shih, & Brennan, 2004). The role that these factors play in the development or maintenance of maternal depression, and resulting impact on youth depression, is not fully understood. A final limitation within the research is the limited number of studies that have examined the effects of fathers with depressive disorders on child outcomes.

Neurobiology

While research has not identified a specific region of the brain that causes depressive disorders to develop, we have observed abnormalities in multiple structures and in the functioning of certain areas of the brain. However, there are few studies in this area that include children and adolescents. Therefore, the application of many of these findings to youth with depressive disorders must be done with caution.

The role of the hypothalamic-pituitary-adrenaline (HPA) axis in depressive disorders has been studied for several decades; however, attention to children and adolescents specifically has only shifted in the past two decades (Lopez-Duran, Kovacs, & George, 2009). Findings within this area have been inconsistent, with some researchers arguing that the role the HPA axis plays significantly differs from adult populations. In general, research has shown there is hyperactivity along the HPA axis in individuals with depressive disorders (Arborelius, Owens, Plotsky, & Nemeroff, 1999). The HPA axis is the neuroendocrine system responsible for the body's respond to stress/demand, which may be a mediator for the development of depression. Specifically, changes to the HPA axis in response to stress, such as childhood trauma exposure, may mediate the development of lifetime prevalence of depression (Heim, Newport, Mletzko, Miller, & Nemeroff, 2008). The role of cortisol and its interaction within the HPA axis has also been a specific area of focus, and recent research suggests that trauma exposure, genetic vulnerability, and female gender all may stimulate sensitivities along the HPA axis in childhood, thereby increasing risk of depression in adulthood (Heim et al., 2008). Importantly, much remains unknown regarding depression and the role of neurobiology and neuroendocrinology, and more research specifically targeting biological markers and replication is needed.

Cognitive Factors

Depression researchers also continue to examine cognitive biases (i.e., negative belief systems, maladaptive information processing) and their role in the development and recurrence of depressive symptoms. In addition to biological markers, the past decade's research has provided significant support for cognitive vulnerability-stress models of depression in youth. That is, when certain cognitive vulnerabilities are paired with specific life stressors, children and adolescents are more likely to develop depressive symptoms (Hammen, Rudolph, & Abaied, 2014). Two primary theories form the building blocks of this relationship: Beck's (1967, 1987) cognitive theory of depression and Seligman's (1975) learned helplessness model. Beck's model identifies three important aspects of cognitive functioning related to depressive tendencies: (1) dysfunctional attitudes and negative cognitive schemas that determine how information is processed, (2) thinking biases that lead to negative interpretations of events, and (3) the "negative cognitive triad" which includes a low or devalued self-concept, an unjust world view, and hopelessness related to the future. Seligman's model originally identified a link between "learned helplessness" and depressive symptoms. Subsequent revisions of this model added the concepts of a "depressive attributional style" (Abramson, Seligman, & Teasdale, 1978) and "hopelessness" (Abramson, Metalsky, & Alloy, 1989).

As with other areas related to depressive youth, developmental pathways are always a consideration. As children develop abilities to analyze their environment, apply logic and reason, and integrate information across time, cognitive vulnerability may also develop. Stressors that occur

throughout adolescence (i.e., increasing autonomy, increasing life stress) may contribute to the development of various aspects of cognitive vulnerability (Rudolph & Hammen, 1999). Further, a review conducted by Lakdawalla, Hankin, and Mermelstein (2007) found that the link between cognitive vulnerability and depression strengthens with age. More research is needed to develop our understanding of the individual features of cognitive vulnerability, how they fit within developmental pathways, and potential processes that may contribute to trait stability.

Symptom Presentation

According to the DSM-5 (2013), the common feature of depressive disorders is "the presence of sad, empty, or irritable mood, accompanied by somatic and cognitive changes that significantly affect the individual's capacity to function" (p. 155). Depression often coexists with other disorders, making its differentiation problematic. Further, in children, emotion does not always present itself in the same ways as it does in adults and may not be as easily recognized. Irritability and aggressive behavior should be primary considerations when diagnosing children with depressive diagnoses. As mentioned, in years past, many medical professionals referred to "masked depression" when treating youth, referring to cases where behavioral or physical symptoms were easily identified, while the cognitive and affective aspects were less identifiable (Harrington, 1993). Normal processing of immense difficulties and the intense emotions that co-occur should be carefully considered in differential diagnosis. Loss and grief may lead to a great deal of suffering and sadness. Substance misuse, medical conditions, and medication side effects should also be assessed for thoroughly.

While expression of symptoms are not categorically mutually exclusive and may overlap organizationally, examination of individual symptom categories may be valuable in diagnosing and treating. Thus, symptom categories of depression as described in the DSM-5 are below: (1) affective, (2) somatic or physical, and (3) cognitive.

Affective

The most commonly identified affective symptom in the adult presentation of depression is a depressed mood or sadness and a diminished interest or lack of pleasure in typically enjoyed activities. As described, in youth, irritability and low frustration tolerance have been identified as markers of depressive expression. In numerous longitudinal studies, children identified as chronically irritable were at increased risk for internalizing symptoms linked to depression (Copeland, Shanahan, Costello, & Angold, 2009; Leibenluft & Stoddard, 2013; Stringaris & Goodman, 2009; Stringaris, Cohen, Pine, & Leibenluft, 2009). Further, a 20-year community-based study was conducted of adolescent-reported irritability and its related outcomes in adulthood (Stringaris, Cohen, Pine, & Leibenluft, 2009). Irritability significantly predicted MDD and dysthymia. In fact, irritability identified with the diagnosis of oppositional defiant disorder has been linked to later depressive disorders (Eley, Lichtenstein, & Stevenson, 1999; Kuny, Althoff, Copeland et al., 2013). Further, persistent negative affect that is related to multiple topics or that is situationally unchanged should be considered in relation to sadness or loss of interest. Identified feelings of worthlessness, excessive or inappropriate guilt, and hopelessness may also be indicative of depressive difficulties.

Physical

A primary DSM-5 symptom expression in both adults and children is an observable lack of or decline in motor activity. This may be the physical manifestation of a depressed or sad mood, a lack of interest or pleasure, fatigue, or a lack of energy. It may present as slowed speech, thinking, or body movements. Speech that is low in volume, inflection, amount, or variety of content can also characterize this depressive expression. While slowed motor activity seems intuitive, agitated psychomotor activity may also be representative of low mood. This may be observed as the inability to sit still, pacing, hand-wringing, or

pulling or rubbing of the skin, clothing, or other objects. Other expressions of a depressed or sad mood may include tearfulness, crying, or whining. Irritability can be observed as temper outbursts, aggression, or dangerous behavior. As mentioned previously, these observations are also primary symptom criteria used to identify disruptive mood dysregulation disorder.

Significant weight loss or a marked, otherwise unexplained, decrease or increase in appetite may also be an identified physical feature of depressive disorders. Often this symptom presents in younger children as a failure to make developmentally appropriate weight gains. Appetite difficulties may emerge as hyperphagia in adolescent or teen populations. Excessive sleep or lack of sleep (i.e., insomnia or hypersomnia) may be apparent as well. Adolescents and teens are more likely to experience hypersomnia as compared to insomnia.

In addition to these difficulties, complaints of physical pain (e.g., headaches, joint, abdominal, or other pains) are common somatic manifestations of depressed states in both adults and youth. Due to youth being less likely to fluently and comfortably express emotional states, somatic symptoms may be among the first observed (van der Veek, Nobel, & Derkx, 2012). Somatic symptoms may also provide information about depressive severity in adolescent populations (Bohman et al., 2010). The creation of specific suicidal plans and suicide attempts is further observable evidence of emotional, physical, and cognitive pain. These and other concerning behaviors may lead to psychiatric hospitalization, perhaps the most unambiguous evidence of depressive difficulties.

Cognitive

A primary cognitive manifestation of depressive disorders includes a diminished ability to think clearly, to concentrate, or to make clear decisions on a daily or near daily basis. Thought-based symptoms may also be identified by slowed speech, thinking, and body movements or noticeable increases in pauses before responding. Negative thoughts in relation to self-concept, such as low self-esteem or lack of confidence, should also be noted. Less equivocal are recurrent thoughts of or about death and recurrent suicidal ideation, with or without a specific plan.

Finally, symptoms may present within friend or family networks, as disruption in relationships and trouble initiating or maintaining them. Without other easily identifiable causes, suffering school performance may indicate consideration. Additionally, an inability or refusal to participate in activities that were once enjoyable or that may be fun to a healthy youth may signify depressive behavior.

Symptom Measures

Valid and reliable assessment instruments help to quantify the presence and severity of depressive symptomatology and should be used to aid in the identification, diagnoses, and treatment monitoring of depression in youth. There are numerous depression screening measures for children and adolescents available for purchase and many that are available for free online. Many of the most used and well-validated instruments have not yet been or are still in the process of being updated for use with the DSM-5 diagnostic criteria, particularly for use with children and adolescents. All instruments should be used in combination with other clinical data. The American Psychological Association has also identified a number of not yet validated instruments that may be used for further research and clinical evaluation with youth. These measures are included in the DSM-5 manual and available to download online.

Comorbidities and Associated Impairments

One important aspect of studying depression in youth populations is accurately isolating the disorder and its symptoms among high levels of comorbidities. Understanding relationships between these comorbidities can facilitate identifying treatment that is most appropriate and will yield the best results for individual clients. Each client's case is unique and may present its own

web of intricate connections with other disorders. Throughout the lifetime, nearly 30% of people are diagnosed with more than one mental health conditions (Kessler et al., 2005). This section outlines some of the key research regarding the disorders that are co-diagnosed with depression most often in children: anxiety disorders, ADHD, persistent depressive disorder, conduct problems, and substance use disorders.

Anxiety Disorders

Research has demonstrated that anxiety often presents conjointly with depression. In fact, estimates of comorbidity of anxiety disorders and depression among youth range from 15% to 75% (Cummings, Caporino, & Kendall, 2014). While anxiety is often diagnosed in younger populations due to its earlier age of onset (Fichter, Quadflieg, Fischer, & Kohlboech, 2010), depressive symptoms are frequently not identified until adolescence due to higher prevalence at that age range (Woodward & Fergusson, 2001). Despite the increase in research regarding this comorbidity, studies do not frequently determine the chronological occurrence of these disorders (i.e., whether they co-occur or one precedes the other; Cummings et al., 2014). Of the anxiety diagnoses, generalized anxiety disorder (GAD) has the most symptoms and risk factors in common with depression (Cummings et al., 2014).

While there are several theoretical models that describe these co-occurring diagnoses within adult populations, there is still a need for models that describe them in youth (Cummings et al., 2014). Two models that have demonstrated some success within this population include the tripartite model (Clark & Watson, 1991) and behavioral inhibition/activation (Gray, 1987, 1991). However, it is suggested that both models lack sufficient consideration of developmental change and the multiple pathways that should be considered in a youth population (Cummings et al., 2014.; Essau, 2003; Olino, Klein, Lewinsohn, Rohde, & Seeley, 2010). Additionally, as with many other comorbid diagnoses, consideration should be given to the overlap in diagnostic criteria.

Attention-Deficit/Hyperactivity Disorder (ADHD)

Children and adolescents diagnosed with ADHD are at an increased risk for meeting symptom criteria for depression at higher rates than nonclinical populations (APA, 2013; Humphreys et al., 2013; Jensen et al., 2001). Humphreys et al. (2013) conducted two related studies seeking mediator variables in this relationship. The first was a cross-sectional sample of 230 5- to 10-year-old children, and the second was a longitudinal sample of 472 youth followed from birth to 20 years. Supporting previous research findings (Barkley, Fischer, Edelbrock, & Smallish, 1991; Deault, 2010; McLeod, Weisz, & Wood, 2007), both studies found parent-child problems significantly mediated the relationship between attention problems and depression more than the academic or peer domains did. Further research is needed to determine specific responsible components within the parent-child relationship. Worth noting, when peer problems were added as a simultaneous mediator, academic functioning was not found to be a significant predictor of the ADHD and depressive relationship.

In another study assessing mediators in the relationship between depression and ADHD within a community sample of 232 children, child cognitions and parenting behaviors were identified (Ostrander & Herman, 2006). Only parent management training mediated the ADHD and depressive relationship for those children under 8 years old. While parent management training continued to mediate the relationship of those children over 10 years of age, the youth's locus of control was also found to be a significant predictor.

Persistent Depressive Disorder (Dysthymia)

According to the DSM-5, individuals with persistent depressive disorder (P-DD) are at higher risk than those with MDD for comorbid diagnoses and at especially high risk for anxiety and substance use disorders (APA, 2013). Of particular

concern for youth populations, early-onset P-DD is strongly associated with the diagnosis of personality disorders. Research regarding this relationship in youth populations is needed.

Conduct Disorder

In their review of comorbidity of depression and conduct problems in childhood and adolescence, Angold and Costello (1993) found that 22.7–83.3% of participants with depression also met criteria for oppositional defiant disorder or conduct disorder. Conversely, 8.5–45.4% of those with oppositional defiant disorder or conduct disorder also met criteria for depression. More recently, Greene et al. (2002) found that 30% of clinic-referred youth diagnosed with major depression also meet the criteria for conduct disorders/problems.

Opinions of clinicians and researchers often differ regarding the development of this relationship (Wolff & Ollendick, 2006). There is some evidence that symptoms of conduct disorder may lead to circumstances in which depression is more likely to emerge (Zoccolillo & Rogers, 1991), while other studies assert that depression is more likely to precede conduct problems (Kovacs, Paulauskas, Gatsonis, & Richards, 1988). Individually, depression tends to increase as children move into adolescence and young adulthood, while conduct problems tend to decrease as children develop; this may explain some aspect of the peak at middle adolescence of the comorbidity of these two disorders (Beyers & Loeber, 2003; Loeber, Russo, Stouthamer-Loeber, & Lahey, 1994). Interestingly, research shows that an earlier onset of comorbidity is associated with aggressive behaviors such as bullying, stealing, and lying, while onset in middle or late adolescence is often characterized with behaviors that demonstrate acting out against authority such as defiance and truancy (Kovacs et al., 1988). Boys have been found to have a more pronounced presentation of this contrast than girls.

Researchers posit various explanations for the high prevalence of this relationship in adolescence. Some suggest that methodological flaws (specifically biases of those involved in referring or treating clients and the methods they employ to do so may contribute to this trend (Angold, Costello, & Erkanli, 1999; Caron & Rutter, 1991). Other explanations focus on common or overlapping risk factors, such as individual, environmental, and biological circumstances that may influence the development of both depression and conduct disorder (Fergusson, Lynskey, & Horwood, 1996; Seligman & Ollendick, 1998).

Lewinsohn et al. (1995) examined a community sample of 1507 adolescents with various stand-alone and comorbid diagnoses. Participants with both a major depressive disorder and a conduct disorder/disruptive behavior were more likely to have academic problems and poor global functioning than participants with only major depressive disorder. Although presentation of harmful symptoms is prominent in this population, research suggests that those with both depression and conduct problems may be less responsive to treatment (particularly medication) than those with other co-occurring diagnoses (Hughes et al., 1990). Further, they may have higher rates of depression recurrence (Rohde, Clarke, Lewinsohn, Seeley, & Kaufman, 2001).

Substance Use Disorders

In their seminal study of prevalence of psychopathology, Lewinsohn, Hops, Roberts, Seeley, and Andrews (1993) evaluated 3896 children aged 9, 11, or 13 at study start. Authors found that while depressive disorders were common in youth from the age of 9, substance use disorders (SUDs) were rarely discovered in young children and become more prevalent during middle childhood (i.e., 12 to 16 years old, on average). In their more recent meta-analysis of internalizing disorders and comorbidity, O'Neill, Conner, & Kendall, (2011) asserted that SUDs are frequently present in those with internalizing disorders. Prevalence rates of comorbidity ranged from 11.1% to 32%. This high level of variability was attributed to differences in experimental designs, including sample origin, gender composition, and the details of the other preexisting mental health disorders.

Personality

With regard to depressive disorders, the most researched personality trait is neuroticism (Eysenck & Rachman, 1967). Neuroticism can be defined as anxiety proneness and is associated with higher reactivity within the limbic system, which was thought to regulate emotional reactivity. A more recent term, negative affect, is used synonymously within the literature to refer to "levels of subjective distress and unpleasant engagement" (Middeldorp et al., 2006). Individuals who score high on neuroticism also exhibit high levels of negative affect. High levels of neuroticism, or negative affect, are associated with increased risk of developing major depressive disorder. In an updated assessment of the tripartite model, Chorpita and colleagues found that a two-factor model best fits anxiety and depression constructs with negative affect significantly, positively correlated with both depression and anxiety, and positive affect negatively correlated with depression only. Further, physiological hyperarousal was positively correlated with panic disorder only, as found in adult samples (Chorpita, 2002; Chorpita, Plummer, & Moffitt, 2000).

Academic and Social Functioning

Children and adolescents with depressive disorders experience symptoms that are likely to impact their intellectual and academic functioning (e.g., difficulty concentrating, loss of interest in activities). Research in this area has demonstrated consistent impairments in attention, information processing, and executive function (Snyder, 2013; Reppermund, Ising, Lucae, & Zihl, 2009; Rock, Roiser, Riedel, & Blackwell, 2014). However, whether the impairments of cognitive functioning are a consequence of the disorder or a preexisting trait that contributes to the development of the disorder is not yet understood (Hammar & Adal, 2009; Reppermund et al., 2009). Studies have found that for many individuals, these impairments have been found to persist beyond acute depressive episodes and some continue to be affected by cognitive impairments after remittance (Bhalla et al., 2006; Reppermund et al., 2009).

In addition to academic difficulties, youth experiencing depressive disorders often suffer from difficulties in their interpersonal relationships (Rudolph, Flynn, & Abaied, 2008; Rudolph, Hammen, & Burge, 1994). Problems with social engagement and willingness to participate in peer-valued activities underlie depression treatment techniques for youth specifically targeted at addressing these deficits (Weisz, Thurber, Sweeney, Proffitt, & LeGagnoux, 1997). The social difficulties that frequently co-occur among youth with depressive disorders are frequently studied as antecedents to the development of depression, though there is some research examining social skill impairments as a consequence of depressive disorders (Agoston & Rudolph, 2013). Due to the frequent disruption of important relationships among youth with depressive disorders, current theories about the etiology and manifestation of depressive disorders incorporate interpersonal factors.

Depression and Suicide

Suicidal thoughts and behaviors represent one of the most concerning areas of impairment among youth with depressive disorders. Major depressive disorder is associated with a high rate of mortality, much of which is driven by high rates of comorbid suicidality and consequent suicidal behavior (Nock et al., 2013). Suicide is the third leading cause of death among adolescents in the USA, and for youth with depressive disorders, the rates of suicidal ideation, planning, and attempts are even higher than other youth with mental health difficulties (Avenevoli et al., 2015). Indeed, while nearly all mental health disorders were predictive of suicidal ideation, a recent nationally representative survey of youth in the USA found that only MDD/dysthymia predicted the development of a suicide plan (Nock et al., 2013). Among children and adolescents, the most common methods of suicide completion involve suffocation (47.4%) and firearms (41.0%) (Centers for Disease Control and Prevention [CDC], 2014).

Suicidal thoughts and behaviors among children and adolescents are a matter of critical concern. Given the strong link between depressive disorders and suicide and the prevalence of suicidal behavior among youth, increasing the availability of effective treatments for depression is an important step in suicide prevention.

Course and Developmental Challenges

The ways in which the expression of depressive symptoms varies throughout early development are numerous. Infants may appear subdued and wary of contact with others (Spitz & Wolf, 1946). Preschool-aged children might engage in violent play or imaginative play that focuses on death or suicide (Luby et al., 2003). Throughout the elementary school years, as children gain greater cognitive abilities and are able to verbally communicate their internal states, children begin to express symptoms more commonly associated with typical characterizations of depression. Most adolescents with depression display nearly identical clinical symptoms as adults with depression. The onset of depressive disorders can occur at any point during development and may develop gradually or suddenly. However, for youth that develops depressive disorders, these most often occur during adolescence, after age 12 (Avenevoli et al., 2015).

With the onset of puberty, the onset of what is known as the "double standard" of depression begins. Prior to puberty, it is well established that there is little to no difference between the sexes in terms of the prevalence of depression (Nolen-Hoeksema & Girgu, 1994; Hammen, Rudolph, & Abaied, 2014). However, as children approach puberty, there is a dramatic increase in the rates at which youth experience depression, as well as a significant difference in the proportion of females to males that are affected. Beginning roughly at puberty and lasting throughout the life span, females are twice as likely as males to develop depression. The most common variables identified that influence gender differences include hormonal contributions, social role expectations, stress, and coping processes (Mash & Barkley, 2003).

The average duration of depressive disorders varies based on diagnoses, with "double depression" or dysthymia/P-DD superimposed with major depressive episodes lasting the longest (Kaminski & Garber, 2002). Little is known about the typical length of depressive episodes in children younger than 6 years old (Luby et al., 2003). For school-aged children and adolescents, the average reported length of MDD ranges from 32 to 36 weeks (Kovacs, Feinberg, Crouse-Novak, Paulauskas, & Finkelstein, 1984; Kovacs, Obrosky, Gatsonis, & Richards, 1997). Based on DSM-5 criteria, persistent depressive disorder (P-DD) requires a duration of at least 1 year to be diagnosed in youth; however, this gives little information about the average duration for P-DD. Current estimates of typical duration are drawn from past studies on dysthymia, with clinical studies reporting a mean duration of 156 weeks (Kovacs et al., 1984, 1997). While this estimate remains informative of the typical duration of P-DD, the impact of the consolidation of chronic major depression and dysthymic disorder into the diagnosis needs further investigation.

Treatment

The main treatment options for major depressive disorder (MDD) and persistent depressive disorder (P-DD) fall into two categories: psychopharmacological and psychosocial interventions. *Psychopharmacological interventions* for childhood depression have largely been extensions of the evolution of psychotropic medications for depression in adults. Tricyclic antidepressants were first developed and utilized to treat depressive disorders in the 1950s and ultimately were prescribed for children beginning in the 1960s (Geller, Reising, Leonard, Riddle, & Walsh, 1999). In brief, tricyclics are so named based on their three-ring structure with attached amines, such as imipramine, amitriptyline, and clomipramine. As with most psychotropic medications that are developed for adults and extended to children, prescription prevalence initially increased dramatically and subsequently reduced due to side effects and other safety concerns as well as the introduction of selective serotonin

reuptake inhibitors in the 1980s. Specifically, tricyclic antidepressants were associated with deaths in youth related to cardiac events (Stark et al., 2006).

As evidence for the potential side effects for tricyclics emerged and a new class of antidepressants were developed (selective serotonin reuptake inhibitors [SSRIs]), prescribing for tricyclic antidepressants drastically reduced (Stark et al., 2006). SSRIs were first introduced in 1986 and appeared to be an answer to the problems with tricyclic antidepressants. SSRIs, as their name suggests, effectively prevent the reuptake of a majority of released serotonin (5-HTP), thus increasing the amount of total serotonin in the synaptic space. Serotonin is critical in the experience of positive mood and the reduction of negative affect, thus having more 5-HTP available for a longer amount of time within the synaptic space, combined with certain genetic markers, appears critical to mood improvement. Examples of the most commonly prescribed SSRIs include fluoxetine (Prozac), sertraline (Zoloft), and citalopram (Celexa; Hughes et al., 1999).

Though the most common antidepressants have been approved by the Food and Drug Administration to be prescribed to youth under 18 years, concerns related to prescribing psychotropic drugs for youth remain (Emslie et al., 2002). Problems with side effects similar to those found in adults, such as problems with appetite and sleep, are also concerns with youth (Stark et al., 2006). Further, research has demonstrated a heightened risk of suicidality, or thoughts and/or attempts of suicide, in youth treated with antidepressant drugs. A meta-analysis by Bridge and colleagues in 2007 revealed a modest but significant elevated risk among pediatric patients (Bridge et al., 2007). In addition to concerns regarding potentially lethal side effects, another problem with the use of psychotropic medications to treat depression in youth is that physicians often prescribe off label. Due to difficulties in studying off-label prescribing, little is known regarding potential beneficial *or* detrimental effects of non-indicated antidepressants in children and adolescents. Finally, caregivers may hold various reservations about psychotropic medications that make them resistant to seeking out pharmacological interventions, including reservations about side effects or religious or cultural beliefs that discourage the use of medications.

Given the concerns and potential side effects related to psychopharmacological interventions, psychosocial treatment is the other frontline option for depressed youth. A recent, updated meta-analysis conducted by Weisz et al. (2013) examined the psychosocial treatment literature for youth and found 52 studies meeting their criteria for comparing psychosocial treatment to usual care. Their findings revealed that the mean effect size was 0.29, with a 58% probability that a randomly selected youth would have a better outcome from being treated with an evidence-based practice (EBP) than usual care. Importantly, research in both psychosocial and psychopharmacological treatments for depression in youth has shown that treatment effects are moderated by severity of the depression, as well as comorbidity and similar complicating factors (Weisz et al., 2013; Asarnow et al., 2009); thus, tailoring evidence-based approaches to individual needs is necessary.

The research evidence base for psychosocial treatment of child depression primarily consists of cognitive-behavioral treatment (CBT) approaches. For instance, Primary and Secondary Control Enhancement Training (PASCET) developed by Weisz et al. (1997) is a well-established, cognitive-behavioral, evidence-based treatment for youth depression. Treatment involves cognitive-behavioral practices addressing depressive thoughts, emotions, and behaviors based on a skill deficit theory; whereby youth are taught with practices that fall either into a primary control domain (skills that youth can utilize to change/impact negative situations that may exacerbate depressive symptoms) or a secondary control domain (skills that youth can utilize to adapt to negative situations in which rumination may exacerbate depression symptoms). An example of a practice that falls within the primary control domain is "problem-solving," whereas an example of a practice that falls within the secondary domain is "cognitive" in which youth are taught to identify and restructure cognitive distortions. More recently, interpersonal therapy for adoles-

cents (IPT-A; Mufson et al., 2004) has become established as an effective psychosocial treatment for youth depression and has demonstrated positive outcomes in comparison to established CBT approaches (Tang, Jou, Ko, Huang, & Yen, 2009; Mufson et al., 2004). Like CBT, IPT/IPT-A is a time-limited approach to dealing with depression, which is typically focused on current difficulties. Within IPT/IPT-A, the emphasis is on improving interpersonal functioning, specifically, by conceptualizing depression symptoms within one or more of four problem areas (grief, role disputes, role transitions, and interpersonal deficits). Then, strategies for addressing the relevant problem areas are taught and rehearsed.

In addition to these more comprehensive treatment protocols, behavioral activation is both frequently a component of existing protocols and stand-alone, evidence-based practice that has demonstrated effectiveness in depression symptoms (e.g., Hopko, Lejuez, Ruggiero, & Eifert, 2003). Behavioral activation (BA) involves a process of encouraging the client to engage in healthy activities to positively impact the client's mood and overall quality of life. The BA process includes developing an individualized menu of activities that are reliably mood enhancing for the client, as well as encouragement and structure to support engaging in the activities and consistent progress monitoring of the impact of the activities on mood. Both case examples and clinical trials have demonstrated positive effects, particularly when BA includes engaging in activities that are physical or involve physical exercise in some way (Lejuez, Hopko, Acierno, Daughters, & Pagoto, 2011; Hopko et al., 2003).

The largest federally funded study to examine psychosocial treatment was the Treatment for Adolescents with Depression Study (TADS; March, Silva, & Vitiello, 2006), which included a head-to-head comparison of evidence-based psychosocial treatment for depression (cognitive-behavioral therapy [CBT]) to medication (fluoxetine [FLX]), medication plus psychosocial treatment combined (COMB), and a placebo (PLC) control. The TADS began in 1999 and was a randomized controlled trial to evaluate both the short- and long-term effectiveness of the different intervention approaches described above, including the combined approach. Results revealed that at both the 12-week and 1-year follow-ups, effect sizes for the COMB condition were higher than *either* CBT or medication alone (FLX), as well as the placebo (PLC) conditions (March, Silva, Vitiello, & the TADS Team, 2006; TADS Team, 2009). In addition, mixed results were found with respect to suicidality risk and SSRIs. Specifically, SSRI usage was a protective factor with respect to suicidal *ideation* (thoughts of suicide); however, SSRI usage was considered a risk factor with respect to suicidal *behavior*. That is, youth who reported suicide attempts were more likely to be treated in the fluoxetine-only condition (Stark et al., 2006). Thus, the importance of promoting the risk/benefits of psychosocial treatment in comparison to psychotropic treatments to youth/families in need is paramount to consumers of mental health treatment having a complete, informed understanding of their treatment options.

Prognosis and Outcome

Current estimates suggest that at least 50% of individuals who experience one major depressive episode (MDE) will have one or more additional episodes (Burcusa & Iacono, 2007). Of those who experience a second episode, approximately 80% will have additional recurrence (Burcusa & Iacono, 2007). Youth with MDD are considered to have an early age of onset (prior to age 30), which comes with an additional elevation in the risk of recurrence. The often recurrent nature of depression intensifies the significant consequences that are associated with the disorder on both an individual and societal level. In a survey of over 3000 adolescents, 90% of those with recurrent depression reported "very much" impairment, such that they felt their work and social interactions were negatively affected (Wittchen, Nelson, & Lachner, 1998). Paired with the fact that MDD and dysthymia are among the costliest health conditions in the USA and that these costs have increased over the past 25 years, the need for youth access to effective treatments to help alleviate the psychological and economic burdens associated with the disorder is

starkly apparent (Greenberg, Fournier, Sisitsky, Pike, & Kessler, 2015).

Major depressive disorder has been established as a highly recurrent disorder; however, outside of this observation, little is known about what factors affect a child or adolescent's risk of relapse. In adult populations, risk factors include suicidal ideation, a history of childhood abuse, and an earlier age of onset (Oquendo et al., 2013). Interestingly, despite the significantly increased risk for females to develop MDD, studies have found mixed results regarding whether female sex is related to an increased risk of relapse.

The increased likelihood that an individual with a history of depression is more likely to reexperience depression than an individual without such a history suggests that the disorder itself may increase the vulnerability of an individual to a recurrent depressive episode. These observations contributed to the popularization of the *kindling hypothesis* as an analogy for the development of sensitivity to stress. The kindling hypothesis suggests that after the experience of major life stress and a depressive episode, an individual is sensitized to life stress on a biological level, such that less stress is required to produce a similar level of depression than the initial depressive episode (Burcusa & Iacono, 2007; Post, 1992). Thus, following the initial depressive episode, an individual is more likely to experience successive depressive episodes after experiencing lesser intensities of life stress. Current research provides some support for the kindling hypothesis, though as results are inconsistent, more studies are needed in this area (Burcusa & Iacono, 2007).

Several factors that can increase the vulnerability of a child or adolescent developing a depressive disorder have been identified. While these factors can elevate the risk that a youth will develop a depressive disorder, they also provide important information about areas to target prevention or protective efforts. The cost of children and adolescents living with depressive disorders is steep, but with access to disorder-specific treatments (such as those described in the above section), we may be able to reduce the likelihood that these youth will experience lifelong difficulties.

Limitations, Gaps, and Future Directions

There are a number of limitations and gaps within the existing literature in childhood depression which primarily center on lack of information availability in specific areas. Indeed, one of the primary issues is a lack of research on depression in young children (Achenbach, McConaughy, & Howell, 1987; Luby, 2010; Measelle, John, Ablow, Cowan, & Cowan, 2005). Little is known regarding unique symptomatology among young children, and the low prevalence of depression in this age group may or may not be an artifact of undetected symptom presentation. Specifically, for children under 6, reports of depression are almost entirely based on caregiver report, despite evidence that many youth are capable of reporting on their own internalizing experiences (Achenbach et al., 1987). In addition, some studies suggest that symptoms that discriminate between depressed and nondepressed youth may vary at different developmental stages (e.g., loss of interest in preschool age youth, social difficulties and crying in school-age youth, etc.; Coster, Leentjens, Lodder, & Verhey, 2005; Frigerio, Pesenti, Molteni, Snider, & Battaglia, 2001; Luby et al., 2003; Measelle, John, Ablow, Cowan, & Cowan, 2005; Wilcox & Anthony, 2004). Importantly, it is unknown if these discriminative symptoms and potential predictors of depressive episodes remain the same with DSM-5 criteria. Given the strong heritability factors in depressive disorders (Waszczuk, Zavos, Gregory, & Eley, 2014), it is reasonable to assume that even very young children may show signs of early onset of the disorder and yet little research exists examining depression in youth under the age of approximately 7 or 8 years old. Indeed, the majority of psychopathology and treatment literature for childhood depression has utilized adolescent samples, which is likely a reflection of the extension of the adult DSM depression symptom criteria to these youth. More research is needed to specifically understand how depression may manifest in young children, as well as the intersection of developmental stages and depressive disorder symptom criteria.

In relation, another important area for future research is regarding the predictive validity of

emotional awareness/knowledge related to depression. Emotional intelligence is an important first step toward developing emotion regulation, which plays a critical role in the development and maintenance of depression symptoms and other emotional and behavioral difficulties such as anxiety (Sánchez-Álvarez, Extremera, & Fernández-Berrocal, 2016). However, relatively little is known about the predictive validity of low emotional intelligence in the development of depression compared to other aspects of emotion regulation, such as poor cognitive control or problem-solving skills (Pagliaccio, Luby, Luking, Belden, & Barch, 2014). Indeed, it is unknown if developing emotional intelligence/awareness is a mechanism for emotion regulation (van Beveren et al., 2016). This research is particularly important for depression treatment in that identifying the causal mechanisms underlying the development of depression symptoms is critical for targeting evidence-based practices. In addition, research on evidence-based practices as well as usual care suggests that developing emotional intelligence (e.g., affect education/psychoeducation on emotions, feelings, identification, awareness, etc.) is often a frequently occurring component of treatment, though little is known about its relative influence alone or in comparison to other treatment components in reducing depression symptoms (Cohen, Mannarino, & Deblinger, 2012; Garland et al., 2010; Kendall, & Hedtke, 2006; Ollendick, King, & Chorpita, 2006).

Another area in which more research is needed is in regard to the role of sleep in childhood depression. The field of sleep science has exploded in recent years, though what remains clear is that there is still much to learn regarding the role of sleep in the risk, development, and maintenance of various psychopathologies (Clarke & Harvey, 2012; van der Zweerde et al., 2016; Palagini, & Rosenlicht, 2011; Walker & van der Helm, 2009). Though sleep difficulties are a potential symptom criterion for a depression diagnosis, symptoms vary (i.e., insomnia vs. hypersomnia), and more research is needed to clarify the role of sleep. In addition, sleep problems may represent an important risk factor related to the "kindling" hypothesis of affective episodes (Post, 1992, 2007). For instance, a longitudinal study taking place in the Netherlands found sleep problems to be a clear predictor of depression recurrence in adolescence, particularly among girls (van Lang, Ferdinand, & Verhulst, 2007). When combined with other comorbidities or stressful life events, the importance of clarifying the complex mediating and moderating relationships is underscored in order to target prevention and treatment efforts.

Conclusion

Childhood depressive disorders are a diagnostic area that is continuing to evolve. Investigative work throughout the past century has greatly clarified our understanding of these experiences within children and adolescents, and our ability to identify these conditions and provide effective services has improved. However, a number of areas of research remain, particularly given the risk for negative outcomes such as suicide. The large number of correlates associated with the development and maintenance of depression, as well as the fact that not all treatments are effective for all youth, suggests that important aspects of depression research remain unknown. In order to accurately predict mood episode recurrence and individualize treatment practices with youth of all ages, research must continue to build upon current knowledge.

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Part V

Internalizing Disorders: Anxiety Disorders

Separation Anxiety and Panic Disorders in Children

8

Alexandra Zagoloff and Gail A. Bernstein

Introduction

Anxiety disorders are some of the most commonly diagnosed psychiatric concerns in children and adolescents (Beesdo, Knappe, & Pine, 2009; Connolly & Bernstein, 2007; Ehrenreich, Santucci, & Weiner, 2008). At the same time, clinicians working with youth must distinguish clinical concerns from developmentally normative, time-limited anxieties. Beesdo and colleagues defined anxiety as pathological "when it interferes with functioning" and becomes "frequent, severe, and persistent" (Beesdo et al., 2009, p. 484). Anxiety disorders in youth are associated with additional comorbid conditions in adulthood, and consequently the early identification and treatment of these disorders are crucial. This chapter will provide an overview of two common anxiety diagnoses: separation anxiety disorder (SAD) and panic disorder (PD). The literature regarding a link between SAD and PD will also be synthesized.

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Separation Anxiety Disorder

Diagnostic History

Previous versions of the DSM (DSM-III, DSM-III-R, DSM-IV, DSM-IV-TR) included SAD in the category of Disorders Usually First Diagnosed in Infancy, Childhood, or Adolescence. Its primary diagnostic criterion is developmentally inappropriate fear surrounding separation from an attachment figure. This criterion must be demonstrated in at least three of the following ways: significant concern during or in anticipation of separation, worry about loss of a loved one, worry about separation as a result of a tragedy (e.g., kidnapping), difficulty leaving the house due to separation concern, difficulty being alone at home, difficulty sleeping alone, nightmares regarding separation, and somatic complaints related to separation. The DSM-III included two examples of separation-related difficulties: reluctance/refusal to attend school and social withdrawal, apathy, sadness, or difficulty concentrating during separation (American Psychiatric Association [APA], 1980).

DSM-5

In the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) (APA, 2013), SAD was moved from the category of

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childhood disorders to the section on Anxiety Disorders. The primary criterion is still developmentally inappropriate fear surrounding separation from an attachment figure. The criterion related to age of onset (previously before age 18 years) has been removed. The criterion related to duration of symptoms has also been modified; the duration must still be 4 weeks or more in children, but greater than 6 months for adults. The final two criteria (impairment and not due to another mental disorder) remain consistent.

Etiology

A review summarized the literature on the relationships among affectionate control, affectionless control, and childhood anxiety disorders (DiBartolo & Helt, 2007). Affectionate control refers to the idea that parents of children with clinical anxiety may demonstrate more frequent controlling behavior but may be as warm as other parents as they try to shield their children from difficulties. Affectionless control, by contrast, suggests that parents may demonstrate more controlling and less warm parenting strategies. The authors evaluated top-down and bottom-up studies – the former include parents with anxiety disorders and their children, while the latter include anxious youths and their parents. Results of the top-down studies suggested that anxious parents engaged in less warm behavior than non-anxious parents, but the results regarding parental control were inconclusive. The bottom-up studies suggested that anxious children had parents who demonstrated more controlling behavior than the parents of non-anxious youths. The two groups of parents did not differ on measures of warmth. Overall, the bottom-up studies were more consistent with the theory of affectionate control, while the top-down studies differed from both theories.

Neuroimaging of youths with anxiety disorders is currently a robust area of investigation. Fear neurocircuitry is linked with the amygdala, although it remains unclear in what way the amygdala is connected with learning processes (Blackford & Pine, 2012). This circuitry also involves the bed nucleus of the stria terminalis and the prefrontal cortex (ventromedial, ventrolateral, dorsomedial, and dorsolateral). The developmental trajectory of anxiety disorders may be partially explained by the different developmental pathways of key areas within the brain – while the amygdala is one of the earliest regions to be fully developed, the prefrontal cortex, which controls higher-order functioning, is one of the last to be fully functioning. More neuroimaging studies of children with anxiety disorders are needed since the majority of data have been collected on adults and it is difficult to determine whether the findings are due to the underlying disease state or a consequence of disease and/or treatment. One study of socially anxious adolescents included continuous scores on a measure of separation anxiety, and higher scores were associated with increased activity in the amygdala (Killgore & Yurgelun-Todd, 2005).

Symptom Presentation

During development, significant changes occur in cognitive and emotional capabilities that impact the extent to which youths can effectively recognize and relay information about their experiences. As a result, appropriate assessment of symptoms should include multiple informants (child, parent, teacher). Presentations in childhood may differ from adulthood (e.g., anger and tantrums during episodes of anxiety). Finally, the assessment of childhood anxiety disorders is complicated by the developmentally appropriate demonstration of fear in response to different situations across developmental periods. Notable examples include the following: "stranger danger" and shyness in infancy, fear of storms and imaginary monsters in childhood, and fear of negative evaluation in adolescence (Beesdo-Baum & Knappe, 2012).

The onset of SAD is typically in childhood with fewer individuals being newly diagnosed in adolescence (Beesdo, Knappe, & Pine, 2009). In one longitudinal sample, all cases of SAD were diagnosed by age 12, and the profiles were simi-

lar for males and females (Beesdo et al., 2009). SAD is typically one of the earliest diagnosed anxiety disorders (along with specific phobia) with rates of approximately 3% in children and 8% in adolescents. As with most anxiety diagnoses, females are more likely to be diagnosed with SAD than males at a ratio between 2:1 and 3:1 (Beesdo et al., 2009). One of the limitations of the current diagnostic system is that individuals must meet multiple criteria to receive a diagnosis. As a result, children may experience subclinical difficulties (with regard to separation or other concerns), not receive a diagnosis (or instead be diagnosed with Anxiety Disorder, Not Otherwise Specified), and as a result not receive treatment and/or not be accounted for in epidemiological research. Nonetheless, such children may experience legitimate distress and impairment.

Youths in the Child/Adolescent Anxiety Multimodal Study (CAMS) often described their symptoms as less impairing than their parents did (Kendall et al., 2010). They might underreport their difficulties due to mistrust of the mental health system, lack of insight, social desirability, or avoidance of anxiety. At the same time, parents might over-report their child's anxiety as a result of their own psychopathology or out of a desire to ensure their child receives treatment (Kendall et al., 2010).

Youths with anxiety disorders often complain of somatic symptoms (e.g., headaches, stomachaches) (Connolly & Bernstein, 2007). It is important to gather information regarding physical complaints before initiating treatment (especially with psychotropic medication) in order to distinguish side effects from symptoms of anxiety. Somatic symptoms are also addressed by and respond to cognitive-behavioral therapy (CBT, described later in the chapter).

Associated Impairments and Comorbidities

The CAMS trial treated youths with three primary anxiety diagnoses (SAD, generalized anxiety [GAD], social anxiety [SOC]) and their combinations. These three disorders were selected due to their high comorbidities with one another and because they were thought to have similar responses to CBT and selective serotonin reuptake inhibitor (SSRI) medication (Compton et al., 2010). The highest percentage of participants (36%) met criteria for all three diagnoses (Kendall et al., 2010). Less than 4% were diagnosed with SAD alone, and an additional 14% were diagnosed with comorbid SOC or GAD in addition to SAD. Children were more likely than adolescents to be diagnosed with SAD plus SOC or GAD. Non-whites were more likely than whites to meet criteria for SAD plus SOC. Fifty-five percent of participants in the CAMS trial met criteria for an additional DSM-IV disorder beyond one of the three principal anxiety diagnoses. It was most common for a child to receive one additional diagnosis, but some children received two, three, or four additional diagnoses. Attention-deficit/hyperactivity disorder (ADHD), oppositional defiant disorder (ODD), and obsessive-compulsive disorder (OCD) were the most common comorbidities. The authors noted that the high percentage of comorbid externalizing disorders is significant since these disorders can impact treatment.

Developmental Course

Different methodological designs present opportunities and challenges when gathering epidemiological data on the course of disorders (Beesdo et al., 2009). Retrospective studies of adults with anxiety disorders are subject to biases of recall. This method may also overestimate the duration of illness since periods of remission may be forgotten. Prospective, longitudinal studies offer strength in evaluating incidence and prevalence rates, as well as rates of stability, remission, and relapse. Unfortunately, they are often more time intensive and therefore costly.

The Netherlands' Tracking Adolescents' Individual Lives Survey (TRAILS) has followed 2230 individuals from preadolescence into adulthood and measured their mental and physical health (Van Oort, Greaves-Lord, Verhulst, Ormel,

& Huizink, 2009). Three assessments were completed between 10 and 18 years of age and evaluated GAD, SOC, SAD, PD, OCD, and major depressive disorder (MDD). Results revealed an initial decrease in anxiety from preadolescence to early adolescence followed by a subsequent increase. Females consistently had higher levels of symptoms. The authors speculated that the transition into high school might contribute to the increase in symptoms from mid- to late adolescence. Alternatively, increasing independence from parents and higher expectations might be contributing factors. Homotypic correlations (one disorder with itself across time) were slightly higher than heterotypic correlations (one disorder with another over time). Additional research is necessary to determine factors that contribute to one disorder remaining consistent across time versus developing into another disorder. Some of the difference between males and females was explained by differences in depression with females demonstrating greater symptoms.

The Great Smoky Mountains Study (Copeland, Angold, Shanahan, & Costello, 2014) similarly presented data from a prospective longitudinal study. Children (N = 1420) from three cohorts (ages 9, 11, and 13 years) were included at baseline and subsequently were interviewed annually until their 16th birthday. They were also interviewed at ages 19, 21, and 25. The prevalence rate for SAD dropped from approximately 4.5% at 9–10 years of age to approximately 1% at 11–12 years. The prevalence continued to decrease until age 19 when it became close to zero. By age 26, 5% of the total sample had met criteria for SAD, and the rate was not significantly different between males and females. Individuals with SAD were at an increased risk of comorbid depressive disorders. However, when looking at functional outcomes (health, financial, interpersonal), individuals with SAD did not have poorer outcomes than those without a history of childhood anxiety.

Ehrenreich and colleagues (2008) have noted several avenues of research remain regarding the developmental course of SAD. First, providers could identify parents with difficulties tolerating their children's normative separation anxiety so that such parents might receive early psychoeducation aimed at preventing the development of SAD. Treatments would also be beneficial for SAD at critical junctures, e.g., transitioning to school, going away to camp, and sleeping at a friend's house. Lastly, specific pathways should be investigated to determine whether certain manifestations of SAD are predisposed to develop into different disorders. For example, if children who excessively check on their loved ones' safety are predisposed to develop OCD, their treatment may benefit from exposure and response prevention. Alternatively, children whose primary symptoms are somatic may be best treated with relaxation and breathing strategies (Ehrenreich et al., 2008).

The transfer of SAD to the section on Anxiety Disorders should allow for increased attention to adult separation anxiety disorder (ASAD) (Bogels, Knappe, & Clark, 2013). This diagnosis is further differentiated as adult-onset ASAD versus childhood-onset ASAD. Data from the National Comorbidity Survey Replication (NCS-R) (Shear, Jin, Ruscio, Walters, & Kessler, 2006) were evaluated pertaining to SAD in childhood and adulthood. The lifetime prevalence rates were 4.1% and 6.6% for children and adults, respectively. More than a third of the cases (36%) with a childhood onset persisted into adulthood. However, the majority of the adults (77%) reported adult-onset SAD. As such, evidence supports the classification into these two presentations (SAD, ASAD); however, prospective studies are necessary to gather data that are not biased due to retrospective recall of childhood. While the symptoms are similar to SAD in childhood, adults with ASAD may be worried about harm befalling their spouse or offspring, and they may consequently experience impairments related to occupational, familial, and social functioning. Investigators are studying ASAD biomarkers, temperamental antecedents, genetics, information processing, comorbidity, and treatment (Bogels et al., 2013).

Treatment

The CAMS was a randomized controlled trial designed to evaluate the relative and combined efficacy of CBT and SSRIs in youth (Walkup et al., 2008). Children and adolescents ($N = 488$) with moderate to severe SAD, GAD, and/or SOC participated in CBT (14 sessions) or received sertraline (up to 200 mg/day), placebo drug, or a combination of sertraline and CBT for 12 weeks. At posttreatment, 60% of the children with CBT alone, 55% with sertraline alone, 81% with combination treatment, and 24% with placebo were much improved or very much improved on the Clinical Global Impression-Improvement scale. Both CBT and sertraline showed equal efficacy and were significantly superior to placebo for the treatment of childhood anxiety disorders, and their combination yielded a significantly superior response compared to monotherapy. All three active treatments may be recommended with consideration of the availability of the specific treatment and the preferences of the family regarding type of treatment, time involved, and cost (Walkup et al., 2008).

Follow-up has been conducted with CAMS participants at 3 and 6 months posttreatment with 80–83% of participants receiving active treatment maintaining their gains. In both monotherapy groups, the percentage of participants rated as "responders" increased from week 12 to week 24 and again to week 36. As a result, combination treatment was no longer superior to monotherapy at either follow-up time. However, combination remained superior to the monotherapies in terms of percent of participants who reached remission (defined as no longer meeting criteria for any of the three primary diagnoses). The monotherapies were similar to one another in terms of remission rates (52% for both at 36 weeks). Similar to the acute outcomes, effect sizes indicated that by 6 months posttreatment, the outcomes of the three groups were similar enough that none of the active treatments were superior to the others. However, the groups differed in terms of participation in non-CAMS treatments during the follow-up period. Participants in the sertraline alone and CBT alone conditions were more likely than those in combination treatment to seek additional services (36%, 30%, and 16%, respectively).

Barmish and Kendall (2005) reviewed nine CBT outcome trials that included parents in the treatment to investigate whether including parents is beneficial. They cited several reasons for including parents in treatment for children with anxiety disorders: parental psychopathology can impact outcome, children are reliant on their environment, including parents might increase generalization of gains, and parents and children can be actively taught about transfer of control – parents setting up contingencies and then children assuming control as their skills develop. Children in the nine studies had met criteria for a variety of anxiety diagnoses (overanxious disorder, SAD, SOC, GAD, PD). Treatments varied in terms of the parental goals but generally covered the following: removing reinforcement of anxious behavior, modeling appropriate behavior, reducing family conflict, psychoeducation, relaxation training, and building social support. Other differences included the format of parental involvement (individual sessions, parent group, parent and child together) and frequency of involvement. Effect sizes for treatments including parents ranged from small to large. Typically, when parents were involved in treatment, they reported that their child had improved. Currently, there is insufficient evidence to conclude that treatment outcomes are superior when parents participate; however, long-term follow-up is necessary to evaluate whether children demonstrate greater progress following parental involvement in treatment. The authors noted that there may be circumstances in which parental involvement is more important. For example, participation may be crucial when the presenting problem is separation anxiety or if the parents meet criteria for an anxiety disorder themselves. Parental participation may be less helpful during adolescence when independence is a goal.

Given the young age of many children diagnosed with SAD, investigators have developed modifications of traditional individualized CBT to address the importance of parental involvement and not-yet-developed cognitive capabilities

of young children. Puliafico, Comer, and Pincus (2012) adapted Parent-Child Interaction Therapy (PCIT) (McNeil & Hembree-Kigin, 2010), an intervention designed for youths with externalizing disorders, to fit the treatment needs of youths with SAD. The treatment begins with child-directed interaction (CDI), including praise, reflection, imitation, description, and enthusiasm. Next, bravery-directed interaction (BDI) includes psychoeducation and exposure instruction for parents. Finally, parent-directed interaction (PDI) includes teaching parents how to effectively give commands and execute separation-related exposures. The CALM Program (Puliafico, Comer, & Albano, 2013) similarly builds on PCIT to address multiple anxiety disorders in young children by training parents to guide their children through exposure situations in session. In CALM, parents learn the following skills to guide their child through exposure: describing the situation, approaching the situation, directly commanding approach behavior, and selectively attending to approach behavior. Both of these modified treatment approaches have preliminary evidence supporting their utilization (Puliafico et al., 2012).

Given the data supporting CBT for the treatment of youths with anxiety disorders, research has transitioned to identifying the mechanisms through which the treatment is helpful, ways in which to increase its efficacy, and novel approaches to dissemination. Regarding mechanisms of change, recent areas of interest include improving emotion regulation abilities (Hannesdottir & Ollendick, 2007), the therapeutic alliance (Chu, Skriner, & Zandberg, 2014), and change in cognitive evaluations (Hogendoorn et al., 2014). Interventions have been adapted to address specific parental concerns (e.g., parent anxiety; Hudson et al., 2014), particular functional impairments (e.g., difficulty attending summer camp; Santucci, Ehrenreich, Trosper, Bennett, & Pincus, 2009), and cultural diversity (Pina, Holly, Zerr, & Rivera, 2014). Finally, in order to reach a larger percentage of children and families needing services, CBT is being implemented in school-based mental health systems (Weist et al., 2014), pediatric primary care (Kolko & Perrin, 2014), and with technological assistance (Jones, 2014)

Pharmacotherapy for SAD and PD

Medications for SAD and PD in children and adolescents will be discussed together in this section. Medications are considered in the treatment of SAD and PD when the symptoms are so severe or impairing that participation in CBT is difficult or when CBT has failed. Medications are also considered as part of a multimodal treatment plan for youths with severe anxiety disorders. Older children and adolescents are more often prescribed antianxiety medication compared with younger children.

Selective Serotonin Reuptake Inhibitors

The first-line pharmacological treatment for youths with SAD or PD is the SSRIs (Connolly & Bernstein, 2007; Kodish, Rockhill, Ryan, & Varley, 2011; Strawn, Sakolsky, & Rynn, 2012). Several randomized double-blind placebo-controlled studies support the efficacy and short-term safety of the SSRIs for treatment of youths with SAD (Birmaher et al., 2003; Research Unit on Pediatric Psychopharmacology Anxiety Study Group, 2001; Walkup et al., 2008). These studies included participants with SAD, GAD, and/or SOC. In the study by Birmaher et al. (2003), participants with GAD and SOC showed significantly greater improvement on fluoxetine compared with placebo. Participants with SAD demonstrated no difference in clinical response on fluoxetine compared with placebo. However, the SAD patients with higher anxiety severity at baseline showed a trend toward greater response on fluoxetine than on placebo.

There is one open-label study (Renaud, Birmaher, Wassick, & Bridge, 1999) and one chart review (Masi et al., 2001) of SSRIs for

PD. Renaud et al. (1999) treated 1 child and 11 adolescents with PD for 6–8 weeks with an SSRI; some participants received a benzodiazepine concurrently. Nine were treated with fluoxetine, two with paroxetine, and one with sertraline. At posttreatment, 75% were rated as much or very much improved on the SSRIs. Further, 67% no longer met PD criteria. Masi et al. (2001) reviewed the charts of 18 children and adolescents who received paroxetine for PD. Eighty-three percent were rated as markedly improved based on retrospective reviews of the charts. Half of the subjects were panic-free after 10 weeks of treatment. The mean duration of treatment was 12 months. Although the findings from these two studies suggest that SSRIs are efficacious for PD in youths, the lack of a placebo control group and lack of blind raters of improvement make the results tentative.

SSRIs are associated with both physical and psychiatric side effects (Strawn et al., 2012). The most common physical side effects include gastrointestinal symptoms, headaches, and insomnia. A common psychiatric side effect, especially in young children, is motor activation. Other psychiatric side effects include behavioral disinhibition, agitation, bipolar switching, and suicidal ideation and behavior.

Atomoxetine

Atomoxetine is a selective norepinephrine reuptake inhibitor. In a study of children and adolescents with ADHD and comorbid anxiety disorders (i.e., SAD, GAD, and/or SOC), youths were randomly assigned to 12 weeks of atomoxetine or placebo (Geller et al., 2007). The atomoxetine group compared with the placebo group demonstrated significantly greater improvement on both ADHD and anxiety symptom rating scales. Overall, atomoxetine was well tolerated. Appetite suppression was the only side effect that was more common in the atomoxetine group compared with the placebo group. These results suggest that children with ADHD plus anxiety disorder may benefit from a trial of atomoxetine.

Tricyclic Antidepressants

Controlled studies of tricyclic antidepressants (TCAs) for SAD and/or anxiety-based school refusal show contrasting findings with respect to efficacy (Berney et al., 1981, 2000; Bernstein, Garfinkel, & Borchardt, 1990; Bernstein et al., 2000; Gittelman-Klein & Klein, 1973; Klein, Koplewicz, & Kanner, 1992). Possible explanations for the conflicting results include small sample sizes, different dosages, comorbid disorders, and lack of control of concurrent psychosocial interventions. TCAs require monitoring of electrocardiograms and blood levels. Potential side effects include dry mouth, constipation, lightheadedness, sedation, and urinary retention. The TCAs are dangerous in overdose. Due to these issues, TCAs are not a first-line treatment for SAD.

Benzodiazepines

Research data are limited with respect to efficacy, tolerability, and safety of benzodiazepines for treating pediatric anxiety disorders. However, they may be considered for short-term treatment of acute impairing SAD and PD symptoms in conjunction with an SSRI while waiting for the benefits of the SSRI. Benzodiazepines may decrease anxiety symptoms enough to initiate exposure treatment for SAD and/or PD. Due to the possibility of physical and psychological dependence (Reinblatt & Riddle, 2007), the benzodiazepines should only be used on a short-term basis and not prescribed for youths with a history of substance abuse. Potential side effects of benzodiazepines include sedation, behavioral disinhibition, and cognitive impairment. Abrupt discontinuation of benzodiazepines can be associated with withdrawal symptoms including anxiety, insomnia, and seizures.

Black Box Warning

All antidepressants, including SSRIs and TCAs, as well as atomoxetine, have a US Food and

Drug Administration (FDA) black box warning. The warning requires that physicians inform families of the small risk of children, adolescents, and young adults developing suicidal ideation and suicidal behavior while taking an antidepressant. In addition, the FDA recommends close monitoring of youths on antidepressants. The black box warning is based on a meta-analysis of 14 studies of antidepressants for youths with major depression and 7 studies of antidepressants for youths with anxiety disorders or OCD (Mosholder & Willy, 2006). The meta-analysis demonstrated that the risk of a serious suicidal event (e.g., suicidal ideation requiring hospitalization, suicide attempt) was significantly higher in youths on antidepressants (4%) compared with those on placebo (2%). There were no completed suicides in the studies that were included in the meta-analysis. When the meta-analysis was repeated using only studies of youths with anxiety disorders, there was no significant difference in incidence of suicidal events. Thus, the small risk of developing suicidal ideation or behavior on an SSRI is less likely in those with anxiety disorders compared with those experiencing major depression (Mosholder & Willy, 2006).

Clinical Approach to Medication Treatment

An SSRI is the first choice for pharmacological treatment of SAD or PD. There are no data to support that one SSRI is more effective than another for the treatment of pediatric anxiety disorders. Factors that guide the selection of a particular SSRI include side effect profile, drug-drug interactions, duration of action, and history of a positive response in a first-degree blood relative with anxiety. For youths with a history of noncompliance with taking medication, fluoxetine offers the advantage of a longer half-life; missing doses is unlikely to result in withdrawal symptoms. The physician should assess for a history of bipolar symptoms and/or family history of bipolar disorder due to the risk of an SSRI triggering bipolar switching.

When initiating an SSRI, the physician should start at a low dosage and gradually increase the dosage while evaluating for efficacy and monitoring for side effects. In young children, using a liquid form of the SSRI allows for delivering very low doses which may reduce the likelihood of side effects. Since SSRIs do not result in full remission of symptoms in about half of children with anxiety disorders (Walkup et al., 2008), consideration should be given to a multimodal approach of SSRI plus CBT. After remission of anxiety symptoms is achieved, it is recommended that the SSRI be continued for approximately 1 year. Subsequently, during a low-stress period (e.g., summer vacation), a gradual taper down and discontinuation of the SSRI can be considered (Pine, 2002). If children manifest relapse of anxiety during the taper down and discontinuation of the SSRI, the medication should be reinstated.

Prognosis and Outcome

Rates of remission and response after 12 weeks of acute treatment have been published for the CAMS project (Ginsburg et al., 2011). The former has been defined as being nearly symptom-free and no longer meeting criteria for initial diagnoses. Response, by contrast, is typically defined as meaningful improvement. CBT led to a response in 60% of participants, while remission rates ranged from 20% to 46% depending on the outcome measure. Sertraline was associated with a response in 55% of participants, while remission rates ranged from 34% to 46%. Combination treatment led to a response in 81% of participants, while remission rates ranged from 46% to 68%. All active treatments were more likely to result in remission than placebo treatment. Medication and psychotherapy were equally likely to lead to remission, and combination treatment was the most likely to lead to remission. Regarding predictors of remission, older youths, those of minority status, and those with higher severity scores at baseline were less likely to reach remission. A more recent analysis of the same data showed that higher levels of

parental strain at baseline were associated with lower likelihood of improvement at the end of treatment (Compton et al., 2014). Response to treatment is important given that untreated childhood anxiety disorders increase the probability of developing depressive and substance use disorders later in life (Beesdo, Knappe, & Pine, 2009).

Panic Disorder

Diagnostic History

The primary diagnostic criterion for PD is recurrent, unexpected panic attacks which include abrupt and intense fear that peaks within minutes and is accompanied by at least 4 of the following 13 symptoms: cardiac changes, sweating, trembling, pulmonary changes, feeling of choking, chest pain, gastrointestinal symptoms, feeling dizzy or faint, changes in temperature, numbness or tingling, feelings of unreality or being detached, and fear of losing control or dying. The DSM-III required a minimum of three panic attacks within a 3-week period (APA, 1980). With the DSM-IV, the time frame was removed since infrequent attacks can cause significant interference. The cognitive components (fear of dying, fear of going crazy) were one criterion (instead of two), and feeling faint was separated from feeling dizzy in DSM-III.

DSM-5

Several changes occurred regarding PD in the DSM-5. First, panic attacks can be included as a specifier to any DSM diagnosis (e.g., SOC with panic attacks and MDD with panic attacks). Second, panic attacks are now described as expected and unexpected. The former occur in situations in which the trigger is identifiable ahead of time (e.g., an attack in a social situation for an individual with SOC). The latter is not connected with an identifiable trigger (e.g., nocturnal attacks that occur while an individual is sleeping). Unexpected attacks are required for a diagnosis of PD, but such individuals may have expected attacks as well. Third, PD is no longer linked with agoraphobia. Agoraphobia is defined as anxiety about two or more of the following situations: public transportation, open space, enclosed spaces, being in lines or crowds, and outside of the home. In the DSM-IV-TR, the two sets of concerns were linked such that the following three diagnoses were possible: PD with agoraphobia, PD without agoraphobia, and agoraphobia without history of PD. In the DSM-5, PD and agoraphobia are two distinct diagnoses.

The DSM-5 Anxiety, OC Spectrum, Posttraumatic and Dissociative Disorder Work Group conducted a thorough review of proposed changes for PD (Craske et al., 2010). It was noted that the 13 common symptoms may not accurately reflect the experiences of cultural minority groups. Consequently, they recommended changing "hot flushes" to "heat sensations" and adding a note that culturally specific symptoms (e.g., ataque de nervios in Latin Americans) may be seen but should not be substituted for the minimum four required symptoms. The cognitive components were lumped together at the end for clarity, and the term "going crazy" received quotation marks to clarify that this is not intended to be pejorative but rather a common colloquial. An additional note indicates that the fear or discomfort can occur from a calm or anxious state. This specification was included to help clinicians and patients distinguish episodes of panic from anticipatory or subsequent anxiety. This differential further helps conceptualize attacks as peaking within 10 minutes once baseline anxiety is separated from the attack. Criterion A (2) from DSM-IV-TR, describing changes following an attack, was modified slightly to increase clarity. In addition to marking it as B, two of the potential consequences ("persistent concern" and "worry") were collapsed into one since they frequently co-occur. Finally, "cardiopulmonary disorders" were added as an example of an exclusionary medical condition to highlight the importance of assessing such conditions.

Although the changes described above arose from a comprehensive review of data, others have critiqued some of the changes (Asmundson,

Taylor, & Smits, 2014). For example, PD and agoraphobia have been unlinked as diagnoses, but they remain connected through their symptoms (specifically, fear of panic is included in the diagnostic criteria for agoraphobia). Unlike DSM-IV, DSM-5 no longer specifies that panic occurs in the absence of real danger. As a result, Asmundson and colleagues expressed concern that the panic attack specifier may be added inappropriately and could contribute to unnecessary treatment. Lastly, these authors noted that the DSM-5 section on etiology of PD emphasizes neurobiological explanations and minimizes the etiological roles of cognitive and behavioral explanations.

Etiology

The neuroanatomical hypothesis of PD (Gorman, Kent, Sullivan, & Coplan, 2000) suggests that the disorder includes similar pathways as those involved in the conditioning of fear in animals. In synthesizing the literature on basic and clinical sciences, the authors argue that the central nucleus of the amygdala and its connections explain the physiological and behavioral symptoms associated with panic. Other brain regions implicated in PD include the parabrachial nucleus (increased respiration), lateral nucleus of the hypothalamus (autonomic arousal), locus coeruleus (norepinephrine release), and paraventricular nucleus of the hypothalamus (adrenocorticoid release). Further, the authors note that important connections between the amygdala and the sensory thalamus, prefrontal cortex, insula, and primary somatosensory cortex may contribute to misattributions that individuals with PD ascribe to bodily sensations.

Klein (1993) initially proposed a link between separation anxiety and panic related to false suffocation. He theorized that some individuals possess a suffocation detection system that erroneously detects insufficient oxygen and triggers a panic reaction. Pine and colleagues (1998, 2000) examined ventilatory physiology in children with anxiety disorders to determine whether ventilatory abnormalities connect separation anxiety and panic disorders. In one study, children with anxiety disorders (n = 18) demonstrated differences from healthy controls (n = 15) consistent with the adult literature on PD: larger minute ventilations, larger tidal volumes, and more variable breathing patterns. However, there were no significant differences in carbon dioxide or respiratory rate. The authors suggested that these findings indicate that anxious children may handle carbon dioxide inefficiently due to faulty neural control (Pine, Cohen, Gurley, Brook, & Ma, 1998). Of note, five of the anxious children (compared with no healthy controls) experienced a panic attack or panic-like symptoms following the challenge procedures. All of the five had a current or past diagnosis of SAD. Participants with SAD (mean age = 9.9 years) were significantly younger than participants with PD (mean age = 13.3 years).

Kossowsky, Wilhelm, and Schneider (2013) evaluated cardiac sympathetic and parasympathetic, respiratory, electrodermal, electromyographic, and self-report variables during a paced hyperventilation activity. Participants included children ages 5–14 years old (49 with SAD, 21 with a different anxiety disorder, 39 healthy controls). Contrary to hypotheses, children with SAD did not demonstrate increased panic symptoms or signs of slowed recovery. However, they demonstrated elevated reactivity in respiratory variability, heart rate, and facial muscle activity. The authors noted that adults with PD often produce findings more significantly different from controls than were found in this study. However, the difficulty with voluntary breathing regulation may nonetheless be a link to the development of PD.

A recent meta-analysis included 25 studies investigating the link between SAD in childhood and the development of additional disorders later in life (Kossowsky et al., 2013). Children with a history of SAD were significantly more likely to develop PD than those without SAD (odds ratio = 3.45, 95% CI 2.37–5.03). However, SAD in childhood was found to significantly increase the risk of being diagnosed with any anxiety disorder later in life (OR =2.19, 95% CI = 1.40–3.42), and the link was not specifically stronger for PD than

other anxiety disorders. A history of SAD was not a risk factor for the development of mood or substance use disorders. The authors noted the link between SAD and future psychopathology is correlational and does not reveal cause and effect since a third variable may be responsible for both SAD and future pathology. However, they also acknowledged that SAD early in life can interfere with the development of successful coping strategies and as such may increase vulnerability to anxiety disorders in adulthood as a result of inadequately developed coping mechanisms.

The TRAILS study has been analyzed to identify predictors of panic attacks (Mathyssek, Olino, Verhulst, & van Oort, 2012). In their sample (N = 1493), 19.8% of participants experienced a panic attack between the ages of 10 and 20, and 1.2% were diagnosed with PD. Female sex and low or middle socioeconomic status were risk factors. Problems with social functioning and symptoms of internalizing disorders at age 10–12 were significant predictors of panic attack by age 17–20. Self-report of cognitive difficulties and symptoms of externalizing disorders were also predictors. The agreement between parent and self-report regarding the predictive ability of social difficulties is important given the typically low concordance rate between informants. The authors note that it will be important for future studies to elucidate the potential link between early interpersonal difficulties and the onset of panic. These results were also significant as it is one of the few studies suggesting a link between externalizing problems and panic. Given the difficulties associated with panic attacks, it is important for investigators of externalizing disorders to give more attention to the development of panic.

Studies are beginning to demonstrate genetic findings with PD (Sakolsky, McCracken, & Nurmi, 2012). Individuals with a first-degree relative with PD are at an increased risk of the disorder, and two specific chromosomes (15q, 2q) have been studied (Sakolsky et al., 2012). Three genes have been implicated: catechol-O-methyltransferase (involved in neurotransmission), neuropeptide S receptor gene (involved in neuronal signaling), and transmembrane protein 132D (function unknown) (Sakolsky et al., 2012). However, while the rate of concordance for PD is higher among monozygotic than dizygotic twins, a significant amount of variance is still attributed to environmental factors (Gorman, Kent, Sullivan, & Coplan, 2000).

Symptom Presentation

Data have been published on the prevalence of PD among clinically referred children (Doerfler, Connor, Volungis, & Toscano, 2007). In a sample of 280 children and adolescents, 35 met criteria for PD (16 of them also met criteria for agoraphobia). Mean ages of onset for PD and agoraphobia were 9.9 and 7.2 years, respectively. All 13 symptoms of panic were represented with palpitations, nausea, trembling, and shortness of breath being most commonly endorsed. Concerns related to internalizing and externalizing behaviors were seen across parent-, teacher-, and self-report questionnaires. The authors highlighted that none of the youths had been referred specifically for treatment of panic, and consequently providers should conduct thorough evaluations even when panic is not mentioned.

Associated Impairments and Comorbidities

Although suicidality is commonly associated with mood disorders, youths with anxiety disorders should also be assessed for suicidal ideation (Rodriguez & Kendall, 2014). Eighty-six youth receiving treatment in a university-based outpatient clinic participated in a study evaluating the relationship between anxiety and suicidal ideation. While GAD and SOC were the most common diagnoses, SAD and PD were the primary diagnoses for ten and three of the children, respectively. Eighty percent met criteria for a comorbid anxiety disorder, and many youth had comorbid depressive or behavioral disorders. While 58% of the youths endorsed some suicidal ideation on a self-report questionnaire, only two youths had a score beyond the clinical cutoff. Importantly, only 10% of parents reported that

their child had a history of suicidal ideation, so it is crucial to directly interview youths about this symptom. The high prevalence of comorbidity in the Rodriguez and Kendall (2014) sample was similarly documented in the Doerfler sample (Doerfler et al., 2007) – 33 out of 35 youth were diagnosed with two or more anxiety diagnoses. SAD and GAD were the most common comorbid disorders, but mood disorders and behavioral disorders were also frequently comorbid.

Developmental Course

Craske, Poulton, Tsao, and Plotkin (2001) conducted a longitudinal study evaluating children from 3 years of age through 18 ($N = 992$) in order to identify predictors of panic and agoraphobia at ages 18 and 21. Emotional reactivity at 3 years of age (as measured through a 90 minute cognitive and motor evaluation) was a significant predictor of panic in males. Individuals with a personal or family history of respiratory illness were also at an increased risk of panic/agoraphobia. Results did not support a link between early separation anxiety and risk for panic; rather, males with low separation concerns at age 3 were more likely to experience panic later in life.

The Great Smoky Mountains Study (Copeland et al., 2014) investigated the prevalence of PD. In middle childhood and early adolescence, the prevalence was zero. The sex difference approached significance, such that from age 15 and on (up to age 26), females were more likely than males to meet criteria. While the prevalence for males was approximately 1% for ages 19–26, the prevalence was approximately 2% for females. By age 26, 4.8% of the sample had met criteria for PD. The difference in prevalence between males and females was not significant ($p = 0.15$), but the rate for males was 3.4%, while the rate for females was 6.3%. Individuals with PD were at an increased risk of comorbid depressive and externalizing disorders.

The US National Comorbidity Survey Adolescent is another epidemiological survey investigating the prevalence and correlates of psychiatric disorders in adolescents (Green et al., 2011). The study included a validation of the World Health Organization Composite International Diagnostic Interview (CIDI) in comparison with the Schedule for Affective Disorders and Schizophrenia for School-Age Children (K-SADS). The sample included 10,148 adolescents ages 13–17 in the continental United States. The prevalence of PD was similar when measured using the CIDI in comparison with the K-SADS (2.4% versus 2.1%).

In a European sample, Wittchen, Reed, and Kessler (1998) showed a slow increase in the incidence of PD through childhood with a steeper increase from ages 13–24. Not only were females more likely to be diagnosed with PD before age 14, but also their incidence curve was steeper throughout adolescence. Beesdo et al. (2009) noted that multiple epidemiological studies have documented a low prevalence of PD in childhood (less than 1%) with a larger prevalence in adolescence (2–3%).

Treatment

Panic control treatment (PCT) (Barlow, Craske, Cerny, & Klosko, 1989) has been adapted for adolescents (PCT-A) and was initially described through two case studies (Hoffman & Mattis, 2000). PCT includes exposure to internal sensations (e.g., shortness of breath), exposure to feared situations (e.g., a mall), psychoeducation about panic, breathing retraining, and cognitive restructuring. The adaptation includes the same core components, but modified much of the language to facilitate adolescent comprehension. For example, the terms "physical," "cognitive," and "behavioral" were replaced with "what you feel," "what you think," and "what you do." The article by Hoffman and Mattis (2000) includes a session-by-session description of all 11 appointments. Following treatment, adolescents reported a reduction in the frequency of panic attacks, a reduction in fear and avoidance, scores on self-report questionnaires within the healthy range, and increased engagement in previously avoided activities. The authors noted that their protocol was further adapted in the following ways: the

importance of including parents as between-session "coaches" was highlighted, technical information regarding hyperventilation was simplified in favor of experiential exercises, and they created a pie chart to help adolescents understand the concept of probability overestimation (i.e., overestimating the likelihood of an improbable event).

PCT-A was evaluated with 21 adolescents (age 13–17) in order to identify key points during treatment for change and factors contributing to improvement (Micco, Choate-Summers, Ehrenreich, Pincus, & Mattis, 2007). Nineteen participants were diagnosed with PD with agoraphobia, while two received diagnoses of PD without agoraphobia. In addition to the treatment components previously described, adolescents completed weekly self-report measures on their symptoms (panic, anxiety, depression). Panic attack frequency reduced between sessions 1 and 2 (following psychoeducation), while anxiety ratings decreased significantly between sessions 4 and 5 (after cognitive restructuring was introduced). Following session 5, adolescents were less likely to engage in probability overestimation and were more likely to perceive themselves as capable of coping with a feared outcome. Adolescents were asked to identify the most helpful component of treatment, and the results were (in decreasing frequency) cognitive restructuring, psychoeducation, breathing retraining, exposure to feared situations, and having someone to talk to.

An intensive form of PCT-A has been created to condense the length of treatment, increase access to services, and increase the involvement of clinicians in exposure (Angelosante, Pincus, Whitton, Cheron, & Pian, 2009). The treatment includes six sessions conducted over 8 days and involves approximately 24 hours of work with the clinician. The first three sessions are similar to PCT-A in content. Sessions 4 and 5 are day-long appointments that include multiple exposure activities in the community (e.g., the mall, public transportation, movie theater). The adolescent and parent then conduct exposure activities independently over the weekend before summarizing treatment on Monday and creating a plan for maintaining and generalizing gains. Angelosante and colleagues review several issues with intensive treatment for clinicians to consider: quickly establishing rapport, planning necessary for 2 full days of exposure, keeping the adolescent's motivation high, limited time for homework completion, and using phone call check-ins to provide accountability post-termination. The intensive format of treatment necessitates attention to two additional issues – some individuals may travel a lengthy distance which requires logistical planning (e.g., lodging, transportation) and finances. While the treatment can be costly, the authors note that expensive but brief treatment can be more cost-effective than lengthier interventions.

One randomized controlled trial has investigated PCT-A (Pincus, May, Whitton, Mattis, & Barlow, 2010). Twenty-six adolescents aged 14–17 were assigned to either PCT-A or wait-list condition (monitoring panic and mood symptoms) followed by active treatment. Twenty-two adolescents completed all 11 sessions and were evaluated at posttreatment as well as 3 and 6 months later. All participants were initially diagnosed with PD with agoraphobia, and 12 initiated PCT-A already taking psychotropic medication (SSRI or benzodiazepine). Participants in PCT-A demonstrated clinically significant improvement on clinician-assessed and self-reported anxiety, depression, and anxiety sensitivity. Improvement in PCT-A was significantly stronger than the control group. The improvements on self-reported outcomes were maintained from posttreatment to 3- and 6-month follow-up, while additional improvement was observed on clinician-assessed severity from posttreatment to 3-month follow-up which was then maintained 6 months post-treatment. Adolescents reported satisfaction with the treatment and described breathing retraining to be particularly helpful. Parents were especially pleased to have learned a "common language" for speaking with their adolescents about panic.

Asmundson et al. (2014) noted that changes in the DSM-5 may contribute to improved access to services. Separating PD and agoraphobia may lower diagnostic thresholds, and thus more individuals will qualify for treatment. Adding panic attacks as a specifier for all disorders should also

encourage clinicians to consistently assess these symptoms and provide treatment even in the absence of a diagnosis of PD.

Summary

In conclusion, SAD represents one of the earliest diagnosed psychiatric conditions in childhood. Adolescents, by contrast, are at greatest risk for the development of PD. The etiology, course, treatment, and prognosis of these disorders (and their relationship with one another) are the focus of strong, active research. While effective treatments are available, ongoing work is necessary to improve dissemination and reach children in need. The importance of prevention, early identification, and treatment is especially important given the numerous ways in which untreated illness increases the risk of future additional forms of psychopathology.

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Generalised Anxiety Disorder

9

Lauren F. McLellan and Jennifer L. Hudson

Introduction

Worry and fear can be normal. However, excessive, uncontrollable worry represents the core diagnostic criteria of generalised anxiety disorder (GAD). Youth meeting criteria typically worry about many areas of life, report physical symptoms associated with worry and as a result experience significant distress, impairment and disability. This chapter presents a review of the diagnostic history of GAD as it relates to changes across revisions of the *Diagnostic and Statistical Manual (DSM)*; it then reviews the available research on the symptom presentation, aetiology, epidemiology, comorbidity and impairment of GAD in childhood and adolescents, with a particular focus on the developmental considerations relevant to this age group. The chapter concludes with a description of available treatments and reviews their efficacy in youth. While the focus is on providing a review of GAD in childhood and adolescents, at times, research on overanxious disorder (OAD), the earlier diagnostic form of GAD in childhood and research on GAD in adults will be drawn on where limited research is available on youth.

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The History of GAD Diagnosis

Categorisation of anxiety disorders, particularly generalised anxiety disorder (GAD), has undergone significant changes across revisions of the *Diagnostic and Statistical Manual (DSM)*. For adults, in DSM-III (American Psychiatric Association [APA], 1980), anxiety neurosis (as it was known in DSM-II) was separated into two categories reflecting the presence or absence of panic attacks. In DSM-III the presence of generalised, persistent anxiety (of duration greater than 1 month) was diagnosed when three of four categories of physical symptoms (motor tension, autonomic hyperactivity, apprehensive expectations and vigilance and scanning) were present without panic attacks. However, this generalised anxiety was recognised only in the absence of other Axis I disorders.

Despite the DSM-III criteria, evidence suggested that worry could be present independently of other disorders (Brown, O'Leary, & Barlow, 2001). As a result, in DSM-III-R (APA, 1987), GAD could be recognised as a discrete disorder. In DSM-III-R, GAD was defined by the presence of excessive or unrealistic worry. GAD was categorised by worry across two or more areas that were unrelated to other Axis I disorders, for example, social threat, physical threat and minor life consequences (Butler, Gelder, Hibbert, Cullington, & Klimes, 1987; Craske, Rapee, Jackel, & Barlow, 1989; Sanderson & Barlow, 1990). DSM-III-R

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also placed emphasis on the physical symptoms associated with worry – requiring the presence of 6 out of 18 symptoms grouped into three broad categories – motor tension, autonomic hyperactivity and vigilance/scanning. Despite changes to the diagnostic criteria for GAD, studies found mixed results as to the reliability of a diagnosis, and there was debate about the ability to distinguish GAD from "normal" worry (Craske et al., 1989) or other anxiety disorders like social phobia or obsessive-compulsive disorder (Barlow, Blanchard, Vermilyea, & Di Nardo, 1986). Poor reliability was thought to particularly stem from the limited definition of the two life areas of worry. As a result, it was proposed that more detailed criteria would maximise the reliability of a GAD diagnosis (Rapee, 1991).

Prior to DSM-IV, anxiety disorders in youth were classified separately to those in adults under the category "Anxiety Disorders of Childhood and Adolescence". In youth, DSM-II identified overanxious reaction (APA, 1968), and by DSM-III-R (APA, 1987), a diagnosis of overanxious disorder could be made. Children were diagnosed with overanxious disorder (OAD) if they frequently displayed four of a possible seven criteria within a 6-month period. These criteria included (1) a pattern of excessive worrying about past behaviour, (2) a pattern of excessive worry about future events, (3) excessive concern about competence across more than one area (e.g. social, academic, athletic), (4) somatic complaints where a physical basis could not be ascertained, (5) an excessive need for reassurance, (6) marked self-consciousness and (7) marked feelings of tension or difficulty relaxing. Similar to the concerns about adult diagnostic categorisation, symptoms of OAD overlapped with other anxiety disorders in youth (Bernstein, Layne, Wiener, & Dulcan, 2004) and were present in typically developing youth (Beidel, 1991; Bell-Dolan, Last, & Strauss, 1990). Reliability in making a diagnosis of OAD was also limited (Silverman, 1987). Thus, significant changes were made in DSM-IV to the diagnosis of GAD for adults and children.

In DSM-IV adults and children with excessive, uncontrollable worry that occurred more days than not for a duration greater than 6 months with associated physical symptoms were together categorised as generalised anxiety disorder. The focus of worry for youth diagnosed with generalised anxiety disorder in DSM-IV was not specific to a situation or event and did not occur within the context of another Axis I disorder (e.g. worry about a panic attack, as in panic disorder, or worry about negative evaluation from others, as in social phobia). Instead, worries were broad ranging and typically focused on concerns for performance, concerns about past events, future events (e.g. natural disasters, pollution or crime), appearance and competence. Physical symptoms could include restlessness, fatigue, difficulty sleeping, irritability, difficulty concentrating and muscle tension. In adults, three or more symptoms were required to be present more days than not for 6 months, while children were only required to endorse a single physical symptom. To meet DSM-IV criteria for generalised anxiety disorder, the worry and/or physical symptoms must cause clinical significant interference and distress, or impairment in important areas of functioning should not be due to the physiological effects of a substance and medical condition or be confined to the features of another Axis I disorder (APA, 1994).

The most significant diagnostic change was that youth with excessive worry were no longer diagnosed with OAD, but instead a diagnosis of GAD could be made for children. Although limited research had been conducted on DSM-III-R OAD before it was incorporated into GAD, as defined by DSM-IV (Werry, 1991), diagnosing GAD in youth reflected the developmental trajectory of the disorder (Andrews et al., 2010). Research investigating the overlap between OAD and DSM-IV-defined GAD is mixed. Diagnosis of DSM-III-R OAD has been shown to overlap almost entirely (93–100% agreement) with DSM-IV GAD using parent and child diagnostic interview (Kendall & Warman, 1996; Tracey, Chorpita, Douban, & Barlow, 1997); however, epidemiological data has found smaller rates of agreement (23.5%; Costello, Egger & Angold, 2003), and longitudinal data suggests that children diagnosed with OAD and GAD may have different long-term trajectories (Bittner et al., 2007).

Other changes to the GAD diagnosis from DSM-III-R to DSM-IV were made to overcome the limitations in categorisation of the disorder. Instead of two or more areas of worry, DSM-IV required that worry about *a number* of life events/activities was "excessive". Worry was considered excessive if the intensity, duration and frequency of the worry were out of proportion to the likelihood or impact of the feared event or activity (APA, 1994). DSM-IV GAD also required that the worry was difficult to control. Lack of control over worry rather than the content of worry was found to differentiate clinical from non-clinical worry (Borkovec, 1994; Craske et al., 1989) and GAD from other anxiety disorders (Brown, Moras, Zinbarg, & Barlow, 1993; Meyer, Miller, Metzger, & Borkovec, 1990). The number of physical symptoms that could be endorsed was reduced from a possible 18 symptoms to only 6. Many of the physical symptoms representing autonomic hyperactivity were dropped, while symptoms of motor tension and vigilance more frequently endorsed by patients with GAD were retained (Marten et al., 1993).

The diagnosis of generalised anxiety disorder in DSM-5 has remained unchanged from that of DSM-IV. Criterion A includes excessing anxiety and worry (apprehensive expectation) about a number of events or activities (such as work or school performance) that occur more days than not for at least 6 months. Criterion B specifies that an individual must find the worry difficult to control. Criterion C indicates that the worry is associated with three (or more) physical symptoms [only one symptom is required for children], with at least some symptoms occurring more days than not for the past 6 months. Physical symptoms include restlessness or feeling keyed up or on edge, being easily fatigued, difficulty concentrating or mind going blank, irritability, muscle tension or sleep disturbance (including difficulty falling asleep or staying asleep, or restless, unsatisfying sleep). Criterion D specifies that the anxiety, worry or physical symptoms cause clinically significant distress or impairment in social, occupational or other important areas of functioning. Criterion E ensures that the disturbance is not attributable to the physiological effects of a substance (e.g. a drug of abuse, a medication) or another medical condition (e.g. hyperthyroidism). Finally, Criterion F indicates that the disturbance should not be better explained by another mental disorder (e.g. anxiety or worry about having a panic attack in panic disorder, negative evaluation in social anxiety disorder, contamination or other obsessions in obsessive-compulsive disorder, separation from attachment figures in separation anxiety disorder, reminders of traumatic events in posttraumatic stress disorder, gaining weight in anorexia nervosa, physical complaints in somatic symptom disorder, perceived appearance flaws in body dysmorphic disorder, having a serious illness in illness anxiety disorder or the content of delusional beliefs in schizophrenia or delusional disorder) (APA, 2013).

Aetiology

Intergenerational Transmission There is unequivocal evidence to support the intergenerational transmission of anxiety disorders (Gregory & Eley, 2011; Hettema, Neale, & Kendler, 2001; Merikangas, Avenevoli, Dierker, & Grillon, 1999). A child of a parent with an anxiety disorder is at greater risk of developing anxiety disorders than a child of a non-anxious parent (Johnson, Cohen, Kasen, & Brook, 2007). The mechanisms by which this transmission occurs are likely to be the result of both genetic heritability and specific familial environments that may increase the child's chances of developing an anxiety disorder such as anxious modelling, overprotective parenting or transmission of threat information and avoidant coping (Eley et al., 2015; Rapee, Schniering, & Hudson, 2009). Twin studies of anxiety in children and adults suggest a moderate degree of heritability, with approximately 30–40% of variance in anxiety symptoms accounted for by genetic factors (Eley et al., 2003; Hettema et al., 2001). A significant amount of genetic overlap is evident across different types of anxiety symptoms in childhood and adolescence (e.g. separation anxiety, social anxiety and generalised anxiety) indicating shared genetic risk.

There is also evidence of shared genetic risk between anxiety and depressive disorders. For example, one study by Silberg and colleagues

(Silberg, Rutter, & Eaves, 2001) showed one distinct genetic factor influenced childhood OAD, adolescent depression and childhood phobic symptoms. Further evidence identifying a shared genetic link between GAD and depression has also been observed in studies of adult psychopathology. In a sample of 2163 female adult twins, Kendler and colleagues showed that GAD and major depression were largely influenced by the same genetic factor (Kendler, Neale, Kessler, Heath, et al., 1992). This finding fuelled debate that GAD should be classified as a mood disorder instead of being considered among the anxiety disorders. In a large longitudinal family study of 3021 14–24-year-olds, Beesdo and colleagues provided evidence against this proposition showing that the longitudinal associations between GAD and other anxiety disorders were stronger than those between GAD and depression. In this study, parental GAD was associated with an increased risk in the offspring for anxiety disorders and anxiety comorbid with depression but not depression alone (Beesdo, Pine, Lieb, & Wittchen, 2010).

In addition to providing information about genetic influence, the twin study design can also provide information about the degree to which environmental influences explain variance in anxiety symptoms. Studies utilising data from twins during childhood indicate that a child's shared environment accounts for a significant amount of variance in anxiety symptoms, that is, parts of the environment that make siblings similar. This finding is in contrast to data from many studies of adult anxiety that tend to show little impact of the shared environment on adult anxiety symptoms. Instead, twin studies of children suggest that both shared *and* non-shared environments (environmental influences that make siblings different, that is, unique individual experiences not shared with the child's sibling) have an impact on the variance in anxiety symptoms. Ehringer and colleagues demonstrated significant shared and non-shared environmental influences on GAD in adolescent twins and their siblings (Ehringer, Rhee, Young, Corley, & Hewitt, 2006). Although a number of other studies have also shown support for significant shared environmental influence in general anxiety or trait anxiety in children (Eley & Stevenson, 1999; Eley et al., 2003), there has been two child twin studies of generalised or trait anxiety where little variance was explained by the shared environment (Eaves et al., 1997*;* Legrand, McGue, & Iacono, 1999*)*.

There has been increasing interest, particular in depression research, in the identification of gene-environment interactions (GxE) to explain the development of psychopathology. A GxE occurs when the environment moderates the genetic influence on a particular trait. Although there are limited studies of GAD in adults and children, one study in adolescent female twins showed that the genetic influence on anxiety symptoms significantly increased for girls who had experienced two or more negative life events (Silberg, Rutter, Neale, & Eaves, 2001). Evidence of interactions between specific genetic markers and related constructs (such as behavioural inhibition and depression) has also shown a similar trend: the presence of a specific marker and negative life events increases the vulnerability towards disorder (Caspi, Hariri, Holmes, Uher, & Moffitt, 2010; Caspi et al., 2003; Fox et al., 2005).

In a recent study utilising a novel children-of-twins design, the heritability of adolescent anxiety was examined by comparing the children of MZ twins and the children of DZ twins (Eley et al., 2015). The results of this study demonstrated a clear environmental pathway, independent of genetics, that explain the association between anxiety in the child and parent. The findings of this study are critical to our understanding of the development of anxiety symptoms as it informs us that even after accounting for the influence of genes, an individual's environment significantly shapes the development of anxiety symptoms. Further, the genes that influenced anxiety in the parents were not associated with the genes that influenced anxiety in adolescence, suggesting that perhaps different genes may exert influence on anxiety symptoms in adolescence and adulthood.

Twin studies can inform us about the degree of heritability of anxiety symptoms but do not identify which specific genes might influence the development of anxiety. Molecular genetic studies, utilising a candidate gene approach, have identified a number of different genes (particularly

serotonin markers) each with a likely small effect that may be associated with an increased risk for anxiety and related disorders (e.g. Evans et al., 1997; Lesch et al., 1996; Schinka, Busch, & Robichaux-Keene, 2004). Recently, the first genome-wide association study of anxious children was published but unable, due to the sample size, to identify any markers at the genome-wide significance level (Trzaskowski et al., 2013). This suggests that there are no common genetic variants with a large effect that contribute to the heritability of anxiety symptoms in children. Instead it is likely that anxiety symptoms are influenced by many genes of small effect.

Temperament and Generalised Anxiety Disorders Perhaps the most widely studied temperament construct that has been associated with an increased risk for the development of anxiety disorders is behavioural inhibition (BI). BI is defined as chronic avoidance of or withdrawal from the unfamiliar (Kagan, Reznick, & Snidman, 1987). Children identified as behaviourally inhibited show distress and a reluctance to participate in novel interactions. They also have a tendency to seek proximity to attachment figures. Although BI shares some overlap with the construct of anxiety, it is generally considered to be a distinct construct that confers increased vulnerability to the development of anxiety disorders. There is strong evidence to suggest that BI is associated with an increased risk for later social anxiety disorder, and accumulating evidence to suggest that children identified as behaviourally inhibited is also at risk for GAD (Biederman, Rosenbaum, Chaloff, & Kagan, 1995; Hudson, Dodd, & Bovopoulos, 2011).

There are a number of other temperament and related constructs such as high negative affectivity, effortful control and poor emotion regulation that may also play a role in the development of GAD in children. Lonigan, Phillips and Hooe (2003) showed that negative affectivity was associated with anxiety and changes in anxiety over a 7-month period, in children and adolescents. This is consistent with studies of adults showing a strong relationship between negative affectivity and GAD (Brown, Chorpita, & Barlow, 1998). Muris and Ollendick (2005) proposed that it was the presence of negative affectivity in combination with low effortful control (inhibitory control, attentional control, low intensity pleasure and perceptual sensitivity) that interacts to increase risk for anxiety and depression, such that the child with high negative affect is unable to effectively regulate these intense and frequent emotions. Support for the notion that children who develop anxiety disorders have difficulties regulating emotions comes from clinical studies of anxious children presenting for treatment. These studies suggest that children with anxiety disorders not only have difficulty regulating fear and worry, but they also demonstrate a more general deficit in the regulation of and understanding of a range of emotions (Hurrell, Hudson, & Schniering, 2015; Southam-Gerow & Kendall, 2000; Suveg & Zeman, 2004).

Parent Factors Two styles of parenting have been primarily associated with anxiety disorders in children: control (overprotection, overinvolvement) and rejection (negative, critical parenting). Although few studies have examined the link between parenting style and the presence of GAD specifically, there is a significant body of research identifying a small but significant relationship between parenting styles and the anxiety disorders (Hudson & Rapee, 2001; McLeod, Wood, & Weisz, 2007). The control variable has been more strongly linked to the development of anxiety disorders, while negative parenting has been consistently linked with depression. There is also some evidence to suggest that parenting styles of overprotection and overinvolvement may occur in response to the child's anxiety or anxious vulnerability. A child who is generally anxious and worried may elicit more help and reassurance from a parent. In turn, however, this additional protection and reassurance limits the child's chances to learn accurate threat and coping information about the situation and hence maintains and may further exacerbate the child's anxiety.

More recently there have been a number of studies examining the longitudinal associations between parenting and child anxiety. Hudson and Dodd (2012) showed that maternal overprotection at age 4 predicted later anxiety symptoms, even after accounting for baseline symptoms. In this

study, another strong predictor of later anxiety was maternal anxiety symptoms. Maternal anxiety, as discussed earlier, increases the child's risk of also developing anxiety. An anxious parent may be more likely to model anxious behaviour and more likely to transmit negative coping and threat information to the child, placing the child at greater risk of developing pathological anxiety (Field & Lawson, 2003; Murray, Cooper, Creswell, Schofield, & Sack, 2007; Murray, Creswell, & Cooper, 2009).

Cognitive Bias Information processing biases play an integral role in the maintenance of anxiety disorders during childhood and adulthood (Ehrenreich & Gross, 2002; Vasey & MacLeod, 2001). Specifically, anxiety disorders in children are characterised by attention and interpretation bias towards threat. For example, children with anxiety are more likely to *interpret* ambiguous situations in a threatening way and more likely to *attend* to threat in their environment. Although the evidence for an interpretation bias in anxious children is quite robust, support for an attention bias is less clear due to numerous non-replications and mixed results. One study specifically examining an attentional bias in adolescents showed individuals with GAD demonstrated a significant bias for negative emotional material compared to non-anxious controls (Taghavi, Dalgleish, Moradi, Neshat-Doost, & Yule, 2003). More recently, Waters, Bradley and Mogg (2014) have provided preliminary evidence that children with GAD show different biases to children with other anxiety disorders. Using a visual probe tasks with angry, neutral and happy faces, children with a diagnosis of GAD showed an attentional bias towards threatening faces, while children with other anxiety disorders show a bias away from threat. If this effect is replicated in future studies, it may help to explain the inconsistent findings observed in the field and help us to understand the unique pathways in the development of GAD in children.

A key criterion in the diagnosis of GAD is the experience of excessive and uncontrollable worry. Worry has been defined as "an anticipatory cognitive process involving repetitive, primarily verbal thoughts related to possible threatening outcomes and their potential consequences" (Vasey & Daleiden, 1994; p.186). A number of adult cognitive models of pathological worry have been developed in an effort to understand the development of GAD and related emotional disorders (Borkovec, Alcaine, & Behar, 2004; Dugas, Gagnon, Ladouceur, & Freeston, 1998; Wells, 1995, 2005). Borkovec et al. (2004) proposed that individuals use worry as a form of cognitive avoidance of fear-provoking imagery. The avoidance in turn maintains the worry through the short-term reduction of the threatening images and associated physical sensations.

Wells (1995, 2009) developed a metacognitive model which focuses on the role of metacognitive beliefs (thoughts about thinking) in the development and maintenance of worry and GAD. In this model, Wells proposed that beliefs about the benefits (positive beliefs) and dangers (negative beliefs) about worry maintain pathological worry. With strong support in studies of adult worriers, this model has also been applied to GAD in children (Ellis & Hudson, 2011). For example, Ellis and Hudson (2011) showed that children with an anxiety disorder (both GAD and non-GAD youth) self-reported higher metacognitive beliefs than children without anxiety disorders. There were no significant differences in metacognitive beliefs about worry in anxious children with and without a diagnosis of GAD. This study provides evidence for the role of metacognitive beliefs in anxiety disorders more generally rather than providing support for a unique role in the maintenance of GAD.

There are a number of other cognitive factors that have been hypothesised to play a role in adult worry, such as intolerance to uncertainty (IU) and a negative problem orientation (Dugas et al., 1998). IU has been defined as a tendency to react negatively to situations of uncertainty. In support of its role in worry development, adults with GAD show greater intolerance to uncertainty than adults with anxiety disorders (Ladouceur et al., 1999). Negative problem orientation is defined as a dysfunctional cognitive set involving the tendency to appraise problems as unsolvable threats and lacking confidence in the ability to solve problems (Dugas, Freeston, & Ladouceur, 1997). Borkovec (1985) noticed that although

high worriers are experts in identifying potential problems, they are ineffective problem solvers as they seem to have difficulty implementing solutions. Adults with GAD have also been shown to have poorer problem orientation compared to moderate worriers (Ladouceur, Blais, Freeston, & Dugas, 1998). There is limited research examining these cognitive variables in samples of children with GAD, yet the emerging evidence suggests that like in adults, these cognitive variables are associated with excessive worry in young people (e.g. Parkinson & Creswell, 2011). For example, in a recent community study of 80 8–12-year-olds, Kertz and Woodruff-Borden (2013) demonstrated that IU was strongly associated with clinical levels of worry (Kertz & Woodruff-Borden, 2013). Similarly others have shown that IU was associated with worry in children and discriminated clinically anxious from non-clinical children and adolescents (Comer et al., 2009; Fialko, Bolton, & Perrin, 2012). In one study of clinically anxious children, Read, Comer and Kendall (2013) showed that IU was uniquely associated with a composite diagnosis of GAD; child-reported IU differentiated children with a diagnosis of GAD from children with another principal anxiety disorder (Read et al., 2013). There seems to be an emerging evidence that these cognitive variables are associated with excessive worry in children. However, given the primarily correlational nature of the research, the extent to which these factors are involved in the development and maintenance of GAD in children remains unclear.

Symptom Presentation and Developmental Considerations

Worry is common in youth. Children as young as 7 years of age report worrisome thoughts (Vasey, Crnic, & Carter, 1994), and approximately 67% of children aged between 4 and 12 years report worrying at times (Muris, Merckelbach, Gadet, & Moulaert, 2000). The types of worries reported by children vary during developmental phases. Research indicates that younger children (4–6 years) worry about separation from parents (Muris et al., 2000) or their physical well-being (Vasey et al., 1994), and older children (age 10–12 years) worry about test performance.

While worry and fear can be normal, the excessive, uncontrollable worry that represents the core diagnostic criteria of GAD means that youth meeting criteria typically worry about many areas of life. Specifically, children who meet criteria for GAD typically worry about social situations (e.g. their appearance, friendships), performance (e.g. academic, sport, musical), natural events (e.g. pollution, crime, war, natural disasters), perfectionism (e.g. not making mistakes or getting in trouble), health (their own, others, death, illness) or family (e.g. divorce, financial strain) (Masi, Mucci, Faville, Romano, & Poli, 1999; Suveg, Jacob, & Thomassin, 2009). As with the worries reported by non-clinical children, developmental differences in youth diagnosed with GAD occur. Pina, Silverman, Alfano and Saavedra (Pina, Silverman, Alfano, & Saavedra, 2002) found that a diagnosis of GAD in children was most often associated with child report of worries about their own health, family, and school and parent report that the child worries about the health of others, interpersonal relationships and school. In adolescents, self-reported worries about health (self and others) and parent report of worries about health (self and others) and family were associated with a diagnosis of GAD (Pina et al., 2002). Both children and adolescents with GAD appear to worry about health and family, while school concerns may be more common in children rather than adolescents.

In GAD, worry is also typically associated with many physical symptoms, especially in children and adolescents (Ginsburg, Riddle, & Davies, 2006; Masi et al., 2004). In fact, Ginsburg et al. (2006) found that children and adolescents diagnosed with GAD reported significantly more physical symptoms (6.9 physical symptoms) than youth with social or separation anxiety disorders (4.8 physical symptoms). Feeling restless, experiencing stomach aches and chills or hot flushes were more commonly reported by anxious youth diagnosed with GAD compared to other anxiety disorders. Interestingly, of the three symptoms endorsed more frequently among youth with GAD, restlessness was the only symptom that appears in the diagnostic criteria for the disorder.

Again, the physical symptoms endorsed by youth with GAD may also differ in line with developmental stages. Pina et al. (2002) found that irritability, trouble sleeping and trouble concentrating were the physical symptoms most commonly reported by children who worried at diagnostic levels. In adolescents, inability to sit still or relax, difficulty concentrating and trouble sleeping were associated with a GAD diagnosis.

Overall, children and adolescents with GAD are highly symptomatic. Masi et al. (2004) found that 50% of all children and adolescents (7–18 years) diagnosed with GAD reported the presence of all 11 assessed symptoms (feelings of tension, apprehension, negative self-image, the need for reassurance, irritability, physical complaints, brooding, sleep disturbances, fatigue, psychomotor agitation and difficulty concentrating). Of these, feelings of tension, apprehension, negative self-image, the need for reassurance, irritability and physical complaints were endorsed by 75% of the sample (Masi et al., 2004). Symptoms were not significantly different between children (7–12 years) and adolescents (13–18 years) or males and females (Masi et al., 2004).

Despite high levels of symptomatology, children and adolescents with GAD are, ironically, often overlooked because their concerns mean that they are motivated to behave and perform appropriately and meet high self-standards. They are typically overly mature (for their age) and highly perfectionistic (Suveg et al., 2009). It is not uncommon for children diagnosed with GAD to be sensitive to criticism because they seek to please others and/or to avoid unfamiliar situations because of the uncertainty surrounding these situations. Furthermore, high levels of self-doubt mean that youth with GAD often seek excessive reassurance (Masi et al., 1999). These symptoms of GAD are often misinterpreted as adaptive qualities, at least until the interference associated with the symptoms becomes pronounced (Suveg et al., 2009). As a result, individuals with GAD, especially children and adolescents, are most likely to present to health professionals with physical or sleep problems (Grant et al., 2005; Wittchen & Hoyer, 2001). This help-seeking trend has an impact on the accurate recognition of the disorder and speed and types of treatment that are recommended.

Prevalence, Age of Onset and Course

Epidemiological research on GAD in youth is limited, being hindered by the removal of OAD and introduction of a diagnosis of GAD for youth. Available data suggests that GAD is less prevalent in youth than it is in adults with lifetime prevalence of 0.8% and 12-month prevalence of 0.5–2% (Canino et al., 2004; Ford, Goodman, & Meltzer, 2003; Wittchen, Nelson, & Lachner, 1998). Prevalence of OAD was typically higher than rates of GAD in youth; 12-month prevalence was found to be approximately 2.9% in 11-year-olds in the community (Anderson, Williams, McGee, & Silva, 1987) and 4.6% in primary care settings (Costello, 1989). The 1-year prevalence of DSM-IV GAD in adults generally ranges from 0.8% to 3.6%, with lifetime prevalence rates between 1.4 and 9.5% (typically between 4.1% and 6.6%) (Carter, Wittchen, Pfister, & Kessler, 2001; Grant et al., 2005; Kessler et al., 2005). While anxiety disorders generally appear to be more prevalent in young females than males (Costello, Mustillo, Erkanli, Keeler & Angold, 2003), equal lifetime (Wittchen et al., 1998) and 12-month prevalence rates for GAD (Ford et al., 2003; Wittchen et al., 1998) have been found in boys and girls.

While the tendency to worry excessively can occur early in life, GAD develops later than other anxiety disorders (e.g. specific phobias or separation anxiety disorder) (Kessler et al., 2005). GAD has a varied age of onset beginning at about age eight (Ford et al., 2003; Kessler et al., 2005), but more commonly reaching diagnostic levels during late adolescents or adulthood (Ford et al., 2003; Kessler et al., 2005). Kessler and colleagues found that the average age of onset for GAD was 31 years. Rates of GAD typically increase until about 45–59 years of age, after which point rates reduce significantly (Kessler et al., 2005).

Some research has found the onset of GAD occurs slightly later for males than females (Beesdo,

Knappe, & Pine, 2009). Research suggests that GAD onset during childhood or adolescents is typically associated with increased symptom severity, greater comorbidity and poorer treatment response compared to GAD that develops later in life. Later-onset GAD has been associated with environmental stressors immediately preceding diagnosis (Suveg et al., 2009).

Developmental considerations play a big part in understanding the age of onset of GAD. Worry is considered to be a cognitive thought process that involves anticipation of future outcomes and consequences that are perceived as potentially threatening (Borkovec, Robinson, Pruzinsky, & De Pree, 1983; Vasey & Daleiden, 1994). Vasey (1993) suggested that according to Piaget's theory of cognitive development, children younger than 7 or 8 years may be able to predict the future and therefore worry. As children's cognitive development becomes more advanced and abstract during middle childhood and adolescences, worries are likely to become more prevalent. Research supports the proposal that young children (even as young as pre-school age) have the cognitive capacity for worry but that the capacity to worry increases in line with cognitive development (Ellis & Hudson, 2010). For example, across two studies, Muris and colleagues found that children with average and below-average intellectual abilities (aged between 3 and 14) all reported worries, but worries were more common in children who passed tasks measuring the cognitive developmental milestones associated with the capacity to worry (i.e. ability to predict the future, anticipate multiple potential outcomes) (Muris, Merckelbach, & Luijten, 2002; Muris, Merckelbach, Meesters, & van den Brand, 2002).

The development of metacognition is also likely to impact the onset of GAD. Research on the development of theory of mind suggests that children as young as 3 years of age have an awareness of their own mental state (see Flavell, Green, Flavell, Harris and Astington (1995) for a review). Studies show that 5-year-olds are aware of their own thoughts but that this awareness increases in 8-year-olds and adults (Flavell, Green, & Flavell, 2000) awareness of thinking as a mental state). Finally, studies have assessed the ability for children, adolescents and adults to understand that thoughts can be difficult to control. Results suggest that 9- and 13-year-old children, adolescents and adults understand that thoughts can be difficult to control, while 5-year-olds do not have this awareness (Flavell, Green, & Flavell, 1998). Overall, these research findings investigating developmental considerations parallel data that the age of onset of GAD is rarely younger than 8 years of age and that the prevalence of GAD increases with age.

Research examining the course of GAD suggests that in children and adolescents, GAD appears to have low to moderate stability (Angst & Vollrath, 1991; Gregory et al., 2007). However, in cases where a diagnosis of GAD is not made at follow-up periods, children and adolescents typically meet criteria for other anxiety disorders, demonstrating a continuity of anxiety psychopathology in general (Beesdo et al., 2009). Like other anxiety disorders, a diagnosis of GAD in youth is also associated with increased risk of later developing depression and other mental health concerns (like substance use or suicide; Gould et al., 1998; Woodward & Fergusson, 2001). The comorbidity associated with GAD is also an important consideration when understanding the course of the disorder in youth.

Comorbidity

GAD is associated with high rates of comorbidity. In fact, comorbidity is more common than not for youth diagnosed with GAD. GAD in youth is commonly comorbid with depression, as was a diagnosis of OAD (Bernstein, 1991; Kashani & Orvaschel, 1990; Masi et al., 1999; Strauss, Lease, Last, & Francis, 1988). Approximately 56% of children and adolescents who meet criteria for GAD will also meet criteria for a depressive disorder (Masi et al., 2004). Similarly, GAD is commonly comorbidity with other anxiety disorders. Approximately 75% of children and adolescents who met criteria for GAD also met criteria for another anxiety disorder (35.7% reported only one other anxiety disorder, and 39.4% reported two or more other anxiety disorders; Masi et al.,

2004). Approximately 21% of youth with GAD also met criteria for an externalising disorder (attention deficit hyperactivity disorder, oppositional defiant disorder or conduct disorder). These rates of comorbidity are mirrored in research on GAD into adulthood, where comorbidity is high, especially with depression (Carter et al., 2001; Grant et al., 2005).

Burden and Impairment

A diagnosis of GAD is associated with marked impairment. Wittchen et al. (1998) showed that GAD in youth affects work, school and household management, leisure activities and social contacts, with the greatest impairment to social contacts. In fact, GAD was associated with more impact on social contacts among adolescents and young adult compared with any other mental health problem. Across domains of impairment, GAD (along with recurrent depressive disorder and obsessive-compulsive disorder) led to the most interference in the month proceeding assessment, with 84%, 61% and 46% of youth diagnosed with GAD reporting that social contacts, leisure and work/school/home management, respectively, were very much impacted (Wittchen et al., 1998).

While GAD in youth is associated with high rates of help-seeking, GAD was the only anxiety disorder where youth only sought help from physicians rather than mental health professionals (Wittchen et al., 1998; Wittchen, 2002). These findings parallel others, where a diagnosis of GAD in youth is associated with high rates of health-care utilisation (Wittchen et al., 1998). Certainly GAD is one of the most common mental disorders seen in primary care settings, yet poor recognition of GAD in general practice means that appropriate treatments are either not provided or access to effective treatment is delayed (Lieb, Becker, & Altamura, 2005; Wittchen, 2002). Despite evidence of the impact of GAD in youth, few studies thoroughly investigate the burden associated with the disorder in children and adolescents (Lieb et al., 2005). Adult research shows that the impairment and disability caused by GAD lead to large direct and indirect costs. Studies indicate that the direct economic cost associated with GAD is high (between approximately $ US 733 and $ US 1208 per patient per 3 months). Direct economic costs increase by about 65% when GAD is comorbid with other mental disorders. In addition, indirect costs associated with things like work absence are estimated at an additional $ US 243 and $ US 416 for adults diagnosed with GAD without and with comorbidity, respectively (Souetre et al., 1994).

The disability, impairment and economic costs associated with GAD, coupled with the secondary impact of poor provision of treatment, demonstrate the significant burden on both individuals and the broader community. While many youth present to primary care settings rather than mental health professionals, a range of psychological and pharmacological treatments are available to treat GAD.

Treatment

Although there are currently no specific guidelines for the treatment of GAD in children, practice parameters and clinical guidelines have been developed more broadly for childhood anxiety disorders (American Academy of Psychiatry; Hudson, Creswell, & McLellan, 2014). Given the strong empirical support for the use of cognitive behavioural therapy (CBT), it is typically recommended as the first line of treatment for children with anxiety disorders. There is also preliminary evidence of the short-term efficacy of selective serotonin reuptake inhibitors (SSRIs) and serotonin-norepinephrine reuptake inhibitors (SNRIs). First, we will provide a summary of the structure and content of CBT for GAD and then provide an overview of the evidence for psychological and pharmacological treatments for GAD.

Cognitive Behavioural Therapy A number of cognitive behavioural programmes have been developed for children with GAD including Brave (Spence, Holmes, Donovan, & Kenardy, 2006), Cool Kids (Rapee et al., 2006), Coping Cat (Kendall & Hedtke, 2006), Take Action (Waters et al., 2014) and Friends (Shortt, Barrett, & Fox, 2001). These programmes were developed for

children with a range of anxiety disorders including GAD, separation anxiety disorder (SAD) and social anxiety disorder (SoAD) and are between 10 and 16 sessions in length. This transdiagnostic rather than disorder-specific approach is the result of the high rates of comorbidity among anxiety disorders during childhood and adolescence. Although GAD can be clearly differentiated in childhood, it is generally understood that there is an underlying construct of anxiety that can be treated within a transdiagnostic protocol (Barlow, 2004). Treatment components differ slightly across these transdiagnostic CBT protocols but typically share common skills such as relaxation, cognitive restructuring, gradual exposure, problem solving and parent training. The principle underlying these strategies is that they directly modify the key processes thought to be involved in maintaining anxiety in children. The programmes have been developed to run individually or in a group of similar aged children. Parents are involved in the treatment of children with GAD, yet the degree and manner of parental involvement varies considerably across different protocols, nevertheless resulting in consistent outcomes. Below we describe common components across transdiagnostic CBT programmes, with particular reference to the Cool Kids programme (Rapee et al., 2006).

Psychoeducation In the first session, the therapist provides the child and his/her parents with education about the nature of anxiety as a normal emotion accompanied by (i) bodily feelings (physiological symptoms), (ii) thoughts (cognitions) and (iii) actions (behaviours). The therapist explains the treatment rationale, that is, the programme is designed to teach strategies to manage these three components of anxiety, specifically, emotional awareness and relaxation to identify and manage physical symptoms, cognitive restructuring to think realistically and problem solving and gradual exposure to reduce avoidance.

Cognitive Restructuring Cognitive restructuring involves identifying, evaluating and challenging negative, unhelpful and worried thoughts. This skill is referred to in the Cool Kids programme as "detective thinking" for children or "realistic thinking" for adolescents. The objective of detective thinking is to look for the "facts" – like a detective or scientist – to elicit realistic thinking. The therapist uses examples to assist the child to first understand the link between thoughts and feelings: the way you think affects how you feel. The child is encouraged to identify their worried thoughts like: "Something bad is going to happen", "If I forget my school books, I will get into trouble" or "I must get it perfect". The therapist then assists the child to challenge these worried thoughts using detective thinking.

The "detective thinking" process involves the following steps: (1) identifying the event/situation causing anxiety; (2) identifying the thought behind the anxiety; (3) evaluating how realistic the thought is, for example, by looking for evidence; (4) considering how realistic the expected consequence is; and (5) identifying a realistic thought to replace the unhelpful anxious thought. The therapist encourages the child to develop a series of questions the child (and the parent) can ask about the situation to help collect evidence. Evidence-finding questions include: "What happened last time when you worried about this?" "What are the facts?" "Has this ever happened before?" "How likely is it 'really' to happen?" Worksheets are used to keep track of the process and can become a useful record in the gradual exposure phase of treatment.

Gradual Exposure Gradual exposure is a key component of CBT for GAD in children and involves the child gradually facing anxiety-provoking situations. First, the child and parents are taught about the role of avoidance in maintaining anxiety. That is, avoiding situations that elicit anxiety limits the child's opportunities for learning accurate, "realistic" information about the situation. By facing situations that elicit anxiety, the child learns that the feared expectations are less likely than predicted. Exposure also teaches the child important coping information. A gradual approach is used to increase the child's compliance and to build mastery.

The child and parents are encouraged to identify a list of the child's main fears and worries in order of severity/interference. Then, starting with the child's smallest fear or worry, a stepladder is

built with a hierarchy of small steps to allow the child to gradually face the situation. For example, a common worry for a child with GAD is that he will forget his schoolbooks. The child fears that if this were to happen, he would get into trouble from his teacher. The child may employ a number of safety behaviours that prevent him from learning valuable information about the situation. For example, he may check his bag at least ten times before leaving the house or on the way to school ask repetitive questions of his mum to make sure he has remembered and to ensure he isn't going to get into trouble. To address this worry, the family identifies a goal to work towards: to be able to go to school without checking or asking questions about schoolbooks. In collaboration with the child and the parents, the therapist designs steps to target the child's feared belief: "I will forget my school books" and "I will get into trouble". The first step may involve only allowing the child to check his bag five times before school and only asking mum two questions on the way to school. The child is asked to repeat this step a few times until it becomes easier and he is ready to move on to a more difficult step. A more difficult step might be only checking twice and asking one question. Before implementing each step in a stepladder, the therapist would encourage the child to complete a "detective thinking" worksheet. This is also useful for the parent to refer to when the child is reassurance seeking.

The therapist and the family work to calibrate the steps so the next step is never too hard or too easy. This typically means several adjustments along the way. It is important to find out the elements that make the situation more or less difficult for the child (e.g. is there a certain day that is hardest or a particular teacher that is perceived as more friendly). Using these elements to produce, or amend, a hierarchy ensures that the process begins with easier steps and gets progressively more difficult in small increments. Eventually, one of the harder steps would involve the child deliberately forgetting his schoolbooks. Doing this kind of step allows the child to learn that what he thinks is going to happen is probably unlikely and even if it does happen he can handle it. A good hierarchy will include tasks that are practical so they can be repeated regularly, relate closely to the goal and allow the child to break the cycle of avoidance and safety behaviours. Rewards are given at the successful completion of each step to encourage compliance. As the parents and children develop experience in designing stepladders, the therapist involvement in design can gradually be reduced.

Problem Solving The aim of problem solving is to assist the child and parents to arrive at alternate, less avoidant and more adaptive solutions to a given situation. Typically children with GAD tend to produce limited solutions to a problem, and they tend to rely on behaviours like seeking help and reassurance rather than thinking about alternative more adaptive solutions. Children and parents are taught to identify a specific problem/situation (e.g. I am worried my friend hasn't responded to my invitation) and brainstorm possible responses to the problem (e.g. check in with her at school, ask her parents). In the brainstorming phase, the child is encouraged to list all possible responses without evaluating them as it can reduce creativity. Once the list of possible responses is complete, the parents and therapist encourage the child to consider the advantages and disadvantages associated with each response, and then choose the most ideal response. An important final step is to consider how effectively the chosen response solved the problem.

Relaxation Training Relaxation training can be used as a strategy to assist the child to manage high levels of physiological arousal in challenging situations. First, the child is encouraged to identify the physical symptoms they experience when they worry. These symptoms can then be used as a cue to implement relaxation techniques. Progressive muscular relaxation (PMR) is a common form of relaxation training in which individuals are instructed to alternate between tensing and relaxing different muscle groups. Several scripts for PMR are available and can help guide the child and parents through these techniques both in session and at home. PMR is also sometimes paired with a second relaxation technique

called controlled or deep breathing (for more information on these skills, see Rapee, Wignall, Hudson, & Schniering, 2000).

Worry Surfing This is a technique used to increase the child's tolerance to feelings of anxiety and worry and to reduce rumination. The child is taught that worried feelings have an end just like a wave, and sometimes rather than trying to fight the wave, you can just ride it out or surf it. There are four steps in surfing worry and the physical feelings of anxiety:

- Recognise the anxious feeling.
- Put the feeling in perspective (it's just a feeling) and identify the task at hand.
- Surf the feeling by concentrating on the detail of the task. The aim is to refocus attention on other aspects of the current situation.
- Reward successful surfing.

Parent Management Importantly, there are a number of parenting strategies that can be taught to enable parents to more effectively manage situations in which the child is anxious or worried. There are a number of common traps that parents may fall into when responding to an anxious child such as providing too much reassurance, jumping in too soon (e.g. being overprotective) or allowing and accommodating avoidant behaviour. To address these issues, parents are encouraged to develop new strategies such as:

(i) Shifting the conversation from always answering the child's reassurance seeking questions to asking them, with the help of detective thinking worksheets.
(ii) Using praise and feedback to shape courageous confident behaviour. Rather than focusing on anxious and worried behaviours, parents are taught to pay attention to the child's courage and independence by using affection and praise.
(iii) Encouraging their child's independence in daily activities.
(iv) Allowing natural consequences. For example, if a child accidentally forgets his schoolbooks, instead of bringing the books to the school for the child, the parent is encouraged to allow the natural consequences of the child's behaviour.

The therapist may set specific tasks for the parents to work on at home that focus on a specific behaviour or situation such as "sitting back when the child starts to worry about making a mistake and not rushing in to reassure". The goal is to gradually increase the parent's competence and confidence in responding in more helpful ways to the child's anxiety.

Treatment Outcome

Randomised controlled trials have consistently shown that CBT is efficacious in reducing anxiety disorders and symptoms in children and adolescents. A systematic review conducted by James, Soler and Weatherall (2005) reported that overall 59% of anxiety-disordered children experience remission following skills-based CBT packages. There is further evidence that the effects of CBT persist over time with long-term (uncontrolled) follow-up studies showing maintenance of effects several years following the completion of treatment (Kendall et al., 2015; Saavedra, Silverman, Morgan-Lopez, & Kurtines, 2010). Although further controlled follow-up studies are necessary to comprehensively evaluate this, the evidence suggests that CBT produces clinically meaningful reductions in anxiety that can be maintained over time.

Until recently, the majority of treatment trials in the field have provided information about the efficacy of CBT for childhood anxiety disorders in general, rather than specifically for GAD. With the availability of increasingly large samples sizes made possible through large multisite studies, the identification of differential recovery rates of children with GAD is possible. In fact, there has been preliminary evidence to support the notion that children with GAD have superior outcomes following CBT compared to children with other anxiety disorders like SoAD (e.g. Manassis et al., 2002). For example, in a study of 1519 children with anxiety disorders,

Hudson & Rapee et al. (2015b) showed that children with GAD showed superior improvement in symptom severity and rates of remission compared to children with SoAD and specific phobia. This would suggest that a transdiagnostic protocol has sufficient potency to bring about significant symptom reduction for children with GAD. In another overlapping sample of 842 children with anxiety disorders (including 425 children with a primary diagnosis of GAD), Hudson & Rapeeet al. (2015b) showed that children with a primary diagnosis of GAD had the highest recovery rates at follow-up (57.6%), with significantly greater remission rates than children with SoAD (30.7%). In contrast, Ginsburg and colleagues in a large multisite study of CBT and SSRIs showed that children with GAD did not differ from children with other anxiety disorders in their rates of remission following treatment (Ginsburg et al., 2011). However, this study is unable to provide recovery rates specifically for CBT, given their study also included SSRI treatment, placebo and a combination of CBT and SSRIs.

Taken together, there is strong evidence that a transdiagnostic approach is working well for children with a primary diagnosis of GAD. Nevertheless, there are still a significant proportion of children who continue to meet criteria for an anxiety diagnosis at the end of treatment. Further developments to enhance treatments for children with an anxiety diagnosis, including GAD, are required.

There have only been a handful of studies that have adopted a disorder-specific approach to the treatment of GAD, and these are predominantly pilot studies and case series (Eisen & Silverman, 1993, 1998; Leger, Ladouceur, Dugas, & Freeston, 2003). Some of these disorder-specific programmes are based on cognitive models of the maintenance of GAD and specifically target factors such as intolerance to uncertainty, negative problem orientation, negative beliefs about worry and cognitive avoidance (Leger et al., 2003; Payne, Bolton, & Perrin, 2011). In line with this, Holmes, Donovan, Farrell and March (2014) published a small randomised controlled trial for the treatment of GAD using a disorder-specific approach. The treatment differed from broad-based approaches in that it focused on the cognitive factors hypothesised to maintain GAD (e.g. negative beliefs about worry, cognitive avoidance). Children receiving the disorder-specific protocol (n = 20) showed significant improvements in anxiety compared to children in the waitlist condition. Approximately 53 percent of children in the treatment group no longer met criteria for GAD compared to no remission in the waitlist condition. Interestingly at the 3-month (uncontrolled) follow-up, an impressive 100% of children no longer met criteria for GAD, yet 50% still met criteria for another anxiety disorder. These figures provide preliminary evidence that while a disorder-specific treatment may lead to promising outcomes for GAD, half of the treated children remain anxious. The high rates of comorbidity among the anxiety disorders need to be a consideration in developing efficacious treatments for anxious children. Future research is needed to compare the efficacy of disorder-specific and transdiagnostic treatments for the treatment of GAD and comorbid anxiety disorders.

The evaluation of the efficacy of pharmacological treatment for GAD has similarly been predominantly transdiagnostic in its approach (i.e. focusing on the treatment response to children with a range of anxiety diagnoses). A large-scale review of high-quality studies testing the efficacy of pharmacological treatments of child anxiety disorders found that 58.1% of participants (compared to 31.5% placebo) responded to anti-depressant medication, particularly selective serotonin reuptake inhibitors (SSRIs; Ipser, Stein, Hawkridge, & Hoppe, 2009). Similarly, Strawn, Welge, Wehry, Keeshin and Rynn (2015) reported an overall moderate effect size (Cohen's d = .62) from nine studies using SSRIs in children with GAD, SoAD or SAD with no increases in abdominal symptoms, suicidality or discontinuation from adverse events. In many of these studies, although the specific recovery rates for children with GAD are not provided, the samples are often comprised of a large proportion of children with GAD. For example, in one study using fluvoxamine, over half of the 128 children (n = 73) met criteria for GAD. In this study, 76% of children

receiving the active drug showed significant improvements in symptom severity compared to 29% receiving placebo (Birmaher et al., 2003). Taken together, these results provide evidence of the short-term efficacy and safety of SSRIs in the treatment of anxiety disorders in children.

There have been a small number of studies specifically examining the effects of SSRIs in children with a diagnosis of GAD. In a small sample of 22 children with GAD, children treated with sertraline (25 mg for week 1 and then 50 mg for weeks 2–9) showed significantly greater reduction in symptoms compared to placebo, with 90% of children on sertraline showing endpoint improvements (compared to 10% in the placebo; Rynn, Siqueland, & Rickels, 2001). In one of the largest pharmacological studies of GAD in children and adolescents (n = 323 aged 6–17 years), Rynn and colleagues (Rynn, Riddle, Yeung, & Kunz, 2007) evaluated a serotonin-norepinephrine reuptake inhibitor (SNRI; extended release venlafaxine) showing significant improvements compared to placebo. The authors concluded that extended release venlafaxine is an efficacious and well-tolerated medication in the short-term treatment of GAD in children and adolescents.

Finally, studies have begun to evaluate the effects of combining CBT and medication to treat childhood anxiety disorders. In a recent large multisite study of clinically anxious children, Walkup and colleagues were the first to evaluate a combined psychological and SSRI (sertraline) treatment for anxious children (Walkup et al., 2008). The combination of CBT and medication produced superior outcomes to CBT alone, medication alone or placebo. In fact, combining treatments produced more than 20–25% greater improvement than either treatment alone. At 3- and 6-month follow-up, combined treatment continued to show enhanced outcomes (Piacentini et al., 2014). These findings provide preliminary evidence that adding medication may significantly enhance the benefits of CBT. Future research is needed to evaluate the efficacy of combination treatment specifically for children with GAD and also to evaluate whether these enhanced effects are observed over the long term.

Conclusion

Like DSM-IV, the newest *Diagnostic and Statistical Manual (DSM-5)* defines GAD in children and adolescents as excessive, persistent and difficult-to-control worry about many areas of life. Worry is associated with physical symptoms and causes significant distress or impairment in functioning. While limited research has focused on the developmental of GAD in childhood specifically, a complex interaction between genetic, environmental and cognitive factors is considered to be important. Although GAD occurs in school-aged children, prevalence increases with age in line with developing cognitive and metacognitive capability. Children meeting criteria for GAD experience a broad range of worries that mirror developmentally relevant concerns throughout childhood and adolescents. The disorder is associated with many physical symptoms, high rates of comorbidity and severe impairment, particularly within the social domain. Psychological treatments for GAD and other anxiety disorders in children are typically based on cognitive behavioural principles and are generally considered effective. Pharmacological treatments for GAD and other anxiety disorders include SSRIs or SNRIs and are about equally effective to CBT packages. While the DSM-5 diagnosis of GAD is unchanged from DSM-IV, this provides an opportunity for much needed research to better understand the aetiology, epidemiology, impact and treatment of GAD in children and adolescents.

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Part VI

Internalizing Disorders: Obsessive Compulsive and Related Disorders

Obsessive-Compulsive Disorder

10

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Obsessive-compulsive disorder (OCD) is a heterogeneous psychiatric disorder that affects approximately 1–2% of youth (Geller, 2006; Zohar, 1999) and is characterized by obsessions (e.g., intrusive thoughts, images, or impulses) and/or compulsions (e.g., repetitive behaviors or mental rituals). The disorder has been associated with lower quality of life in youth (Lack et al., 2009) and increased functional impairment in various domains of life (Piacentini, Bergman, Keller, & McCracken, 2003; Piacentini, Peris, Bergman, Chang, & Jaffer, 2007; Valderhaug & Ivarsson, 2005). Given these deleterious effects, a comprehensive understanding of the disorder is necessary to better target the symptomology. Additionally, there are developmental considerations in place when treating pediatric OCD, necessitating more nuanced investigations. As such, this chapter delineates the diagnostic history, etiology, symptom presentation, clinical characteristics, course and prognosis, and treatment of pediatric OCD.

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Initial Overview of Diagnostic History

Unlike many diagnoses that have seen considerable revisions in their diagnostic criteria over the last 30 years, the diagnostic criteria for OCD have remained largely unchanged from its description in the *Diagnostic and Statistical Manual* – III edition (American Psychiatric Association [APA] 1980). However, some changes, which generally reflect increased understanding of the diversity of presentation of the disorder, have occurred across iterations. The following outlines the progression of the diagnostic criteria from the DSM-III up to the most recent criteria set forth in the DSM-5.

DSM-III Beginning in the DSM-III (American Psychiatric Association, 1980), OCD was listed as an anxiety disorder and was defined by three primary criteria: A) the presence of either obsessions or compulsions; B) the symptoms result in significant distress or interference; and C) the symptoms are not the result of another mental disorder. Obsessions were defined as "recurrent, persistent ideas, thoughts, images, or impulses that are ego-dystonic, that is, they are not experienced as voluntarily produced, but rather as thoughts that invade consciousness and are

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experienced as senseless or repugnant" (p. 234). In addition, the diagnostic criteria required that individuals make efforts to ignore or suppress these obsessions. Compulsions were defined as "repetitive and seemingly purposeful behaviors that are performed according to a certain set of rules or in a stereotyped fashion" (p. 234) that are used to either produce or prevent a future event but either do not realistically connect to that desired event or are clearly excessive. Further, it was specified that individuals would experience a sense or urge, despite an initial desire to resist, acknowledge the compulsions to be senseless (except in some young children), and not derive pleasure from the compulsion other than the reduction in distress it provides.

DSM-III-R The DSM-III-R (American Psychiatric Association, 1987) marked one of the larger revisions in diagnostic criteria for OCD but generally maintained the structure set forth by the DSM-III. Regarding obsessions, rather than describing obsessions as ego-dystonic, the terms "intrusive" and "senseless" were introduced. The definition of an obsession was also expanded to include caveats qualifying that in addition/in lieu of efforts to ignore or suppress obsessions, individuals may also engage in compulsions as a method of neutralizing the obsessions. Language specifying that individuals should perceive obsessions as "products of their own mind, not imposed from without" (p. 245) was also added, in order to help differentiate OCD from thought disorders. In addition, the new diagnostic criteria specified that for an individual to have OCD, the obsessions could not be related to the content of another present Axis I disorder, replacing the generic "not due to another mental disorder" (p. 234; American Psychiatric Association, 1980) criteria used in the DSM-III. Information added to the definition of compulsions included introducing the word "intentional" to describe behaviors, introducing language recognizing that individuals may perform compulsions to prevent/reduce current anxiety/distress (rather than just future events), and removing language specifying that the individual does not derive pleasure from the behavior. Finally, in addition to significant distress or impairment in functioning, the DSM-III-R added that individuals could meet diagnosis if time spent on obsessions or compulsions was over 1 h per day.

DSM-IV and DSM-IV-TR With the DSM-IV (American Psychiatric Association, 1994) came additional changes to the diagnostic criteria set forth for OCD. Obsessions now had to be considered as intrusive and inappropriate (rather than senseless) only for "some time during the disturbance" (p. 422) and must cause "marked anxiety and distress" (p. 422). In addition, in an effort to aid in distinguishing OCD from generalized anxiety disorder, it was specified that the obsessions could not be simply "excessive worries about real-life problems" (p. 422). Regarding compulsions, mental acts were also included in addition to repetitive behaviors. Further, language was adjusted so that compulsions were described as something "the person feels driven to perform" (p. 423) rather than as "purposeful and intentional" (p. 423). Criteria for differentiating obsessions from other DSM diagnoses were revised to state that the content of the obsession could not be restricted to another diagnosis, rather than simply related to that diagnosis. In addition, the insight criteria were adjusted to only require individuals to recognize the behavior as unreasonable or excessive "at some point during the course of the disorder" (p. 423). An option to specify the diagnosis for an individual with poor insight was also included at this time. Finally, consistent with other diagnoses in the DSM-IV, a final criterion states that OCD must not result directly from a medical condition or substance. No changes were made between the DSM-IV and the DSM-IV-TR (American Psychiatric Association, 2000).

Presentation of Current Diagnostic Criteria

DSM-5 The diagnostic criteria for OCD set forth in the DSM-5 remain a relatively simple set of criteria. Diagnosis continues to be based on the presence of obsessions and/or compulsions at clinically significant levels, whether due to time

spent, associated distress, or impairment. Compared to the DSM-IV, criteria in the DSM-5 have removed some of the restrictive specifications that had been previously introduced. First, while generally implied in the previous version, specific wording in the DSM-5 now reflects that individuals may present with both obsessions and compulsions, rather than using "either/or" language. Wording of obsessions has been slightly modified, with "impulses" being replaced by "urges" and "inappropriate" being replaced by "unwanted." Further, the specification that obsessions "cause marked anxiety or distress" (which was added in DSM-IV) was specified to occur "in most individuals." The criteria that obsessions must not simply be about real-life problems and that the individual should recognize that the obsessions are thoughts from their own mind (rather than external insertions) are no longer included in the DSM-5. These two criteria are still relevant to OCD but have now been included under the differential diagnosis criteria (i.e., "the obsessive-compulsive symptoms are not better explained by another mental disorder;" p. 237), along with differential diagnosis from other conditions such as stereotypic movement disorder, addictive disorders, and schizophrenia spectrum and other psychotic disorders, which were not explicitly stated in previous versions of the DSM. Importantly, hoarding disorder is now reclassified as its own disorder and is no longer subsumed under the OCD diagnosis. In addition, in the DSM-5, individuals are no longer required to have ever recognized their behavior as excessive or unreasonable. Instead, the specification for insight added in the DSM-IV has been expanded to differentiate between clients with good or fair insight, poor insight, or absent insight/delusional beliefs. Finally, a specification has been introduced to differentiate patients whose symptoms display overlap with a tic disorder. Readers are directed to Leckman et al. (2010) for a thorough discussion of the reasoning and evidence behind these changes.

The largest, most discussed, and debated change to OCD brought forth in the DSM-5 is moving the disorder out of the anxiety disorders and into a new category of obsessive-compulsive and related disorders, which also includes body dysmorphic disorder, excoriation disorder, hoarding disorder, and trichotillomania (e.g., Hollander, Braun, & Simeon, 2008; Mataix-Cols, Pertusa, & Leckman, 2007; Phillips et al., 2010; Stein et al., 2010; Storch, Abramowitz, & Goodman, 2008). Prior to publication, the debate over classification covered a great deal of factors, including genetic and biological predictors, comorbidity, symptomology, treatment response, and expert consensus. Proponents suggested that evidence indicates a stronger relationship between OCD and the disorders that now fall on this spectrum versus the relationship between OCD and anxiety disorders (e.g., Hollander et al., 2008), while criticism against establishing a new diagnostic spectrum focused primarily on the preliminary nature of current evidence and lack of consensus (e.g., Storch, Abramowitz, & Goodman, 2008). While now independent from the anxiety disorder category, this new obsessive-compulsive spectrum directly follows the anxiety disorder spectrum as an effort to reflect the connection that also exists between these categories of disorders (American Psychiatric Association, 2013). This change in classification does not particularly impact the manner in which OCD is diagnosed, but may aid in improving clinician awareness of the relationship between these conditions, in order to improve diagnostic screening and differential diagnosis (Hollander et al., 2008).

Etiology

Genetic and Neurological Obsessive-compulsive disorder has a well-established heritable component with higher concordance rates in dizygotic over monozygotic twins (see van Grootheest, Cath, Beekman, and Boomsma (2005) for a review) and increased risk among first-degree relatives (e.g., Grados, Walkup, & Walford, 2003; Nestadt et al., 2000). Heritability appears to be present across obsessive-compulsive symptom dimensions (Katerberg et al., 2010), and a genome-wide study conducted by Davis et al. (2013) estimated the heritability of OCD to be 37%, with a genetic correlation of 0.41 between

OCD and Tourette syndrome. Given the individual heterogeneity of OCD presentation, the influence of comorbid conditions, and environmental contributions, it is unclear and constituent indicators remain elusive; however, a broad range of genetic and neurological contributors have been examined. Regarding genetic markers, specific links have been suggested (although to varying degrees), with varying impacts (e.g., increased versus decreased risk; sexually dimorphic effects) and with varying consistency for a number of genes, including: glutamate transport gene SLC1A1 (Arnold, Sicard, Burroughs, Richter, & Kennedy, 2006; Stewart, Fagerness, Platko, Smoller, et al., 2007; Stewart, Mayerfeld, Arnold, Crane, et al., 2013); glutamate receptor gene GRIK2 (Sampaio et al., 2011); serotonin transport gene SLC6A4 (Hu et al., 2006; Lin, 2007; McDougle, Epperson, Price, & Gelernter, 1998); 5HT1Dβ (serotonin) receptor gene (Mundo et al., 2002); MAOA gene and COMT enzymatic activity (Karayiorgou et al., 1999); and OLIG2 gene (involved in regulation of white matter development; Stewart, Fagerness, Platko, Smoller, et al., 2007), among others (e.g., Hall, Dhilla, Charalambous, Gogos, & Karayiorgou, 2003; Mattheisen et al., 2014; Stewart, Yu, Scharf, Neale, et al., 2013; Zuchner et al., 2009). Similarly, a range of neurological contributors, including chemical, biological, anatomical, and immunological differences, have been suggested in the pathogenesis of OCD, such as involvement of serotonin and dopamine systems (Barr, Goodman, Price, McDougle, & Charney, 1992; Goodman et al., 1990; Thoren et al., 1980; Zohar & Insel, 1987); dysregulation of glutamate, particularly within the cortico-striatal-thalamo-cortical (CSTC) circuitry (Ahmari et al., 2013; Pittenger, Bloch, & Williams, 2011; Ting & Feng, 2008; Wu, Hanna, Rosenberg, & Arnold, 2012); white matter abnormalities (Bora et al., 2011; Garibotto et al., 2010; Gruner et al., 2012); altered caudate nucleus size (Robinson et al., 1995; Scarone et al., 1992), along with additional pathology within the basal ganglia, orbitofrontal cortex, and thalamus (Brambilla, Barale, Caverzasi, & Soares, 2002; Insel, 1992; Lacerda et al., 2003; Modell, Mountz, Curtis, & Greden, 1989; Rauch & Savage, 1997); and association with infectious triggers (e.g., streptococcal infections; Murphy, Kurlan, & Leckman, 2010; Murphy, Petitto, Voeller, & Goodman, 2001; Swedo, Leckman, & Rose, 2012; Swedo et al., 1998).

Cognitive Behavioral Approach The cognitive behavioral approach to the etiology and maintenance of OCD is based on principles of learning theory, including classical and operant conditioning, and also incorporates ideas from cognitive mediation theory. From a classical conditioning perspective, a previously neutral stimulus becomes conditioned with a distress response. Individuals' cognitive appraisal of the situation can significantly contribute to the initial conditioning of the stimulus. For example, experiencing occasional intrusive thoughts (e.g., "I could crash my car into the median") is common; however, while most individuals simply dismiss the thought as nonsensical, individuals with OCD experience the thought with apprehension believing that the existence of the thought may increase the likelihood of engaging in the behavior (i.e., thought-action fusion; Shafran & Rachman, 2004). Other cognitive distortions common to OCD that increase the likelihood of a fear response include overestimation of threat (e.g., contracting a disease from touching a door handle is highly likely), intolerance of uncertainty (e.g., even if it is unlikely, something bad could happen), inflated personal responsibility (e.g., if something bad does happen, it will be my fault), and perfectionism (e.g., things have to feel just right; Frost & Steketee, 2002). In response to the fear evoked by the newly conditioned stimuli, individuals increase their avoidance and/or develop behaviors (i.e., compulsions) intended to neutralize the threat. Both avoidant and compulsive behaviors temporarily reduce distress, negatively reinforcing both the conditioned nature of the stimulus and the use of these behaviors in future situations. This cycle of negative reinforcement prevents individuals from learning that distress will naturally habituate without avoidance of the stimuli or compulsions, precluding their ability to learn more adaptive responses. Over time, as additional stimuli become paired

with negative responses, individuals become increasingly reliant on avoidant and compulsive behaviors to reduce distress, maintaining the symptomology in the long run.

Symptom Presentation

Obsessive-compulsive symptoms can manifest in a wide variety of ways across individuals; however, amalgamation of patient data has identified that symptoms tend to cluster in a series of unique dimensions. Using responses from symptom checklists, various factor/principal component analyses have been performed in adult and pediatric samples, in an attempt to specifically identify and label symptom dimensions. While these analyses may suggest small dimensional differences, taken as a whole, it appears that OCD symptom expression falls into either three or four categories (excluding hoarding, which has been reclassified as a unique disorder in the DSM-5; Bloch, Landeros-Weisenberger, Rosario, Pittenger, & Leckman, 2008; Mataix-Cols, Rauch, Manzo, Jenike, & Baer, 1999; Stein, Andersen, & Overo, 2007; Stewart, Rosario, et al., 2007). First, analyses have similarly identified a symmetry dimension which includes obsessions surrounding symmetry (e.g., need for things to be just right) as well as compulsions of repeating, ordering, and counting. Second, a contamination/cleaning dimension includes obsessions regarding contamination (e.g., fear of germs), accompanied by compulsive cleaning and washing. Differences have arisen across analyses in the specific combination of the remaining obsessive-compulsive symptoms. Some analyses have suggested a dimension consisting of sexual (e.g., fear of fondling a child)- and religion-based obsessions (e.g., fear of offending God), typically accompanied by reassurance behaviors and mental compulsions (Bloch et al., 2008; Mataix-Cols et al., 1999; Stewart, Rosario, et al., 2007). Reassurance seeking can occur through repeated questioning (e.g., "I am not a bad person, right?"), and mental compulsions commonly manifest as compulsive praying or excessively detailed reviews of past events. In one case, this dimension also included aggressive (e.g., fear of stabbing someone) and somatic (e.g., fear of illness) obsessions, as well as checking (e.g., verifying door locks) compulsions, making a "forbidden thought" category (Bloch et al., 2008); however, others have suggested aggressive/somatic obsessions and checking compulsions function as their own dimension (Mataix-Cols et al., 1999; Stein et al., 2007).

Specific to pediatric populations, checking compulsions appear to co-occur more commonly with symmetry obsessions, while somatic and aggressive obsessions map with the contamination/cleaning dimension (Bloch et al., 2008; Stewart, Rosario, et al., 2007). While the grouping of these dimensions helps identify different manifestations of OCD, individual differences still remain. Some individuals may present with symptoms that clearly are restricted to one dimension, many symptoms across multiple dimensions, or a combination of symptoms that do not fit clear dimensional categorization. Collectively, youth often present with multiple obsessions and compulsions, presenting with rates that are significantly higher than those observed in the adult OCD population (Geller, Biederman, Faraone, Agranat, et al., 2001).

Clinical Characteristics

Functional Impairment Youth with OCD often experience interference in their daily functioning, making it challenging for these children to function optimally across social, familial, and academic domains of life (Piacentini, Peris, et al., 2007; Valderhaug & Ivarsson, 2005). Indeed, increased OCD symptom severity is associated with greater functional impairment (Nadeau et al., 2013; Storch, Larson, Muroff, Caporino, et al., 2010), with more severe youth reporting lower quality of life and physical/psychosocial health, as well as higher difficulties with emotional functioning (Lack et al., 2009). Even after controlling for OCD symptom severity, insight into the symptomology, family accommodation, and depressive symptoms each emerged as unique predictors of OCD-related functional impairment in the child

(Storch, Larson, Muroff, Caporino, et al., 2010). As such, it is apparent that the severity of OCD symptomology and related sequelae both yield deleterious effects on the youth's functioning.

When assessing functional impairment in pediatric OCD, the strength of the link between OCD symptom severity and functional impairment may vary based on the respondent (i.e., youth self-report versus parent report). As such, it is important to consider who the respondent is as well as potential moderators of agreement, such as age, symptom severity, resistance and control over symptoms, and insight (Storch, Rudy, Wu, Lewin, & Murphy, 2014). Additionally, a nuanced examination of the child's presenting obsessive-compulsive symptom dimensions may also yield differential links to impairment. Specifically, the contamination/cleaning and aggressive/checking dimensions have been correlated with impairment in various domains of life, suggesting the expansive impact of these particular symptom clusters (Storch, Larson, et al., 2010). These youth may engage in extensive avoidance and spend an inordinate amount of time worrying and engaging in compulsions, resulting in suboptimal performance across various domains of life.

Comorbidities Youth presenting with an exclusive diagnosis of OCD are relatively uncommon, given that approximately 57–77% of youth with OCD typically present with at least one comorbid psychiatric disorder (Geller, Biederman, Stewart, Mullin, Farrell, et al., 2003; Ivarsson, Melin, & Wallin, 2008; Storch, Merlo, et al., 2008). Specifically, affected youngsters often present with comorbid internalizing and externalizing disorders, including mood, anxiety, disruptive behavior, attention deficit hyperactivity, and tic/obsessive-compulsive spectrum disorders (Geller, Biederman, Griffin, Jones, & Lefkowitz, 1996; Janowitz et al., 2009). There are various factors that may influence the probability of having certain comorbidities, including age at onset of OCD, gender, and length of illness. In particular, individuals with lower age onset have a higher probability of presenting with anxiety, somatoform, impulse control, eating, attention deficit hyperactivity, and obsessive-compulsive spectrum (especially tic) disorders, while having a longer duration of OCD decreased the probability of having a comorbid tic disorder (de Mathis et al., 2008; Geller, Biederman, Faraone, Bellordre, et al., 2001; Hemmings et al., 2004; Janowitz et al., 2009; March & Leonard, 1996; Masi et al., 2006; Millet et al., 2004). Additionally, younger children are more likely to have oppositional defiant disorder when compared to their older counterparts (Ivarsson et al., 2008; Selles, Storch, & Lewin, 2014), and chronological age was associated with a higher prevalence of mood and psychotic disorders (Geller, Biederman, Faraone, Agranat, et al., 2001; Geller, Biederman, Faraone, Bellordre, et al., 2001). Females are more likely to present with comorbid anxiety (especially generalized anxiety disorder), eating, and impulse control disorders (de Mathis et al., 2008; Ivarsson et al., 2008), while males tend to present with comorbid attention deficit hyperactivity and tic disorders (Geller, Biederman, Faraone, Agranat, et al., 2001; Ivarsson et al., 2008).

Attention deficit hyperactivity disorder (ADHD) and disruptive behavior disorders (DBD) have garnered particular attention in pediatric OCD. With ADHD, the inattentive symptoms can be especially difficult to differentiate from obsessional concerns and subsequent distraction, but investigations have indicated that they are disparate disorders that can uniquely coexist (Geller et al., 2002; Geller et al., 2004). Youth with OCD and comorbid ADHD tend to present with a higher frequency of males, earlier onset of OCD, increased psychosocial impairment, and heightened comorbidities with bipolar, tic, and disruptive behavior disorders (Masi et al., 2006). Similarly, youth with OCD and comorbid DBD present with higher family accommodation, OCD symptom severity, impairment, internalizing symptoms, and decreased symptom resistance (Storch, Lewin, Geffken, Morgan, & Murphy, 2010). As such, youth with OCD and comorbid ADHD and/or DBD may demonstrate more convoluted clinical presentations, potentially warranting higher levels of care.

Comorbidities in youth with OCD have potential implications for treatment response. In youth receiving cognitive behavioral therapy (CBT), children with at least one psychiatric comorbidity exhibited lower treatment response and remission rates when compared to youth with no comorbidities (Storch, Merlo, et al., 2008). Indeed, the number of comorbid diagnoses was negatively related to treatment outcome, with ADHD and DBD associated with attenuated response rates, and DBD and major depressive disorder related to decreased remission rates (Storch, Merlo, et al., 2008). When considering response to paroxetine, youth with psychiatric comorbidities demonstrated higher rates of relapse and significantly lower rates of treatment response (Geller, Biederman, Stewart, Mullin, Farrell, et al., 2003). Similarly, tics were found to moderate treatment outcomes in youth taking sertraline, but not in youth undergoing CBT (March et al., 2007). Taken together, psychiatric comorbidities in youth with OCD evidently impact their clinical presentation and pharmacological/psychotherapeutic treatment responses, highlighting the importance of careful assessment and treatment of comorbidities.

Developmental Considerations The presentation of OCD in younger and older samples may differ depending on various demographic and clinical variables. There tends to be a higher preponderance of males in younger cohorts and those with an earlier age of onset (Tukel et al., 2005), and comorbid ADHD occurs more frequently in boys (Geller, Biederman, Faraone, Agranat, et al., 2001). However, when considering early childhood-onset OCD (i.e., age of onset prior to 8), the gender discrepancy does not exist (Garcia et al., 2009). While the severity of OCD symptoms does not appear to differ when considering younger versus older children, younger youth tend to display poorer insight and attenuated ability to resist and control OCD symptoms, while older youth have more intense obsessive-compulsive symptoms (Garcia et al., 2009; Geller, Biederman, Faraone, Agranat, et al., 2001; Selles et al., 2014). Additionally, youth with an early childhood onset of OCD commonly present with a family history of OCD (Garcia et al., 2009). As mentioned above, earlier age of onset for OCD is also associated with various comorbidities, complicating the clinical presentation of these youth.

The phenomenological presentation of OCD may also be distinct when considering various age groupings. When considering chronological age, children displayed higher hoarding symptoms when compared to adults, though sexual obsessions were significantly lower in children (Geller, Biederman, Faraone, Agranat, et al., 2001; Selles et al., 2014). Religious obsessions were more prevalent in adolescents (Geller, Biederman, Faraone, Agranat, et al., 2001), and older youth displayed more magical and somatic obsessions, as well as compulsions related to checking, counting, and superstition (Selles et al., 2014). With regard to age of onset, an earlier age of onset (operationalized as prior to age 17 for the following results) has been associated with increased superstitious/magical, contamination, symmetry/exactness, religious, hoarding/saving, repeating, counting, and tapping/rubbing symptoms (Millet et al., 2004; Tukel et al., 2005). Additionally, early childhood onset (prior to 8 years of age) has been associated with frequent endorsements of obsessions related to aggression and catastrophic cognitions, checking compulsions, and contamination symptoms (Garcia et al., 2009). As such, there may be a phenotypic difference in obsessive-compulsive symptomology when considering age of onset and chronological age.

Course/Prognosis

To date, few studies have detailed the long-term course and prognosis of pediatric OCD. Extant literature has documented the persistence of OCD, with 41% to 68% still experiencing clinical symptoms of OCD into early adulthood and beyond (Bloch et al., 2009; Boileau, 2011; Flament et al., 1990; Micali et al., 2010; Stewart et al., 2004). Various clinical factors have been associated with the persistence of this illness, including female gender, earlier age and higher

OCD symptom severity at the baseline childhood assessment, greater duration of illness, and use of inpatient services (Bloch et al., 2009; Micali et al., 2010; Stewart et al., 2004). However, age of onset has shown conflicting results, as both a later age of onset (Bloch et al., 2009) and an earlier age of onset (Stewart et al., 2004) have both been supported as predictors of persistent OCD. Comorbidities at the baseline assessment (especially oppositional defiant disorder) predicted persistence of OCD, worsened prognosis, and presence of other comorbidities later on (Bloch et al., 2009; Micali et al., 2010; Stewart et al., 2004). Poor initial treatment response for OCD generally predicted poorer prognosis as well (Stewart et al., 2004; with the exception of Flament et al. (1990), where initial response to clomipramine did not predict long-term outcome), with approximately half of youth still receiving treatment or desiring further treatment in early adulthood (Palermo et al., 2011).

When considering quality of life longitudinally, young adults with non-remitting OCD displayed impairment most commonly in the interpersonal and work realms of life (Palermo et al., 2011). Additionally, youth exhibiting primary hoarding symptoms predicted poorer quality of life in adulthood, while the OCD symptoms associated with forbidden thoughts predicted improved quality of life in early adulthood. With regard to obsessive-compulsive symptomology in general, nearly half of individuals with OCD exhibit washing or checking symptoms at some time during the course of illness. However, symptoms typically present from a variety of symptom clusters (Geller, Biederman, Faraone, Agranat, et al., 2001; Rettew, Swedo, Leonard, Lenane, & Rapoport, 1992), with individuals rarely presenting with the same cluster of symptoms longitudinally. Additionally, there appears to be more waxing and waning of symptoms at the microlevel (i.e., the specific obsessive-compulsive symptoms may change within a relatively short time period), though the natural course of OCD broadly appears to be more stable (A. H. Zohar, 1999).

Treatment of Pediatric OCD

Exposure and Response Prevention (ERP) For mild-to-moderate cases of OCD, ERP is the gold standard treatment for pediatric OCD (American Academy of Child and Adolescent Psychiatry, 2012) and is recommended in the care of all patients regardless of severity (American Academy of Child and Adolescent Psychiatry, 2012; National Collaborating Centre for Mental Health (NCCMH), 2006). In this behavior-based variant of CBT, youth confront their feared stimuli and consequently refrain from engaging in their compulsions. This intervention is purposed to target and break the aforementioned negative reinforcement cycle that maintains the OCD symptomology (Kircanski, Peris, & Piacentini, 2011; Lewin, Wu, McGuire, & Storch, 2014), allowing the youth to confront their feared situations without needing to engage in maladaptive compulsions. Exposure and response prevention has consistently demonstrated significant decreases in OCD symptomology in affected youth, exhibiting effects greater than placebo, pharmacotherapy, and control comparison groups (Barrett, Healy-Farrell, & March, 2004; Piacentini et al., 2011; Storch, Geffken, Merlo, Mann, et al., 2007; The Pediatric OCD Treatment Study (POTS) Team, 2004; Watson & Rees, 2008). Long-term follow-ups demonstrated the durability of ERP in maintaining treatment gains, as measured up to 18 months following the termination of acute therapy (Barrett, Farrell, Dadds, & Boulter, 2005; Barrett et al., 2004; Piacentini et al., 2011; Storch, Geffken, Merlo, Mann, et al., 2007).

Exposure and response prevention is conducted in a structured, controlled manner. A fear hierarchy is collaboratively constructed, and it serves as a blueprint for conducting gradated exposures. The anxiety-provoking stimuli are ranked in order from least distressing to most distressing, with the goal of starting with easier exposures and titrating upward. When conducting exposures, it is important to allow the youth to habituate to the anxiety and/or learn to tolerate the distress (Abramowitz, 2013; Arch & Craske, 2011) before moving onto harder exposures.

Once milder exposures are repeated, prolonged, and mastered, the therapist will ask the child to move onto confronting stimuli that provoke higher levels of anxiety. Exposures can be conducted in vivo or imaginally, depending on the feared situation. While the former type of exposure is preferred, certain types of obsessions may be more difficult to implement in vivo (e.g., obsessions related to religion or sex), necessitating the utilization of imaginal exposures (Abramowitz, Braddock, & Moore, 2009). By confronting the feared stimuli, youth are able to learn that their feared consequences are unlikely to occur, discouraging the need to perpetually engage in their compulsions.

In a typical implementation of a course of ERP, psychoeducation about OCD and the maintaining factors are provided to the family (similar to other CBT protocols). Thereafter, a brief overview of various cognitive strategies is introduced. Cognitive tools are primarily used to help externalize the disorder, motivating the child to adaptively handle the symptomology. A typical cognitive tool is identifying thinking traps, such as catastrophizing ("If I touch this contaminated pencil, I'm going to contract a disease"), overresponsibility ("I have to pick up that piece of trash, or else somebody may fall and die"), and thought-action fusion ("If I think that my mom will get into a car accident, she will get into a car accident;" Amir, Freshman, Ramsey, Neary, & Brigidi, 2001; Shafran & Rachman, 2004). By labeling and recognizing the OCD symptoms, the youth is encouraged to build confidence and resist giving into the obsessions and compulsions. However, clinicians must use this technique cautiously, as cognitive restructuring can inadvertently morph into a compulsion (e.g., excessive self-reassurance), necessitating clinical finesse when employing these techniques. Thereafter, the core of treatment is devoted to gradated ERP, as described above. For instance, a child with contamination fears may be asked to touch a "contaminated" surface (e.g., doorknob), sit with the anxiety until it naturally habituates and/or the child learns to tolerate the distress, and ultimately refrain from engaging in handwashing compulsions. Once that task has been repeated and mastered, exposures will move to slightly more difficult tasks, proceeding gradually up the fear hierarchy. At the end of ERP, a review of the therapeutic skills is conducted and relapse prevention is implemented.

Pharmacotherapy For moderate to severe cases of pediatric OCD, medication is indicated as a supplement to ERP, with selective serotonin reuptake inhibitors (SSRIs) considered as the frontline medication for OCD (American Academy of Child and Adolescent Psychiatry, 2012). As a monotherapy, a meta-analysis investigating various SSRIs revealed significant decreases in OCD symptomology that were greater than placebo comparisons, achieving a pooled standardized mean difference of 0.46 (Geller, Biederman, Stewart, Mullin, Martin, et al., 2003). While pharmacotherapy can be well tolerated, dosage titration should be achieved slowly, and potential side effects should be considered (Murphy, Segarra, Storch, & Goodman, 2008), particularly for younger children (Martin et al., 2004). Additionally, for youth experiencing partial response to serotonin reuptake inhibitors, they may achieve greater treatment gains if CBT is concurrently administered, compared to youth continuing solely with pharmacotherapy (Franklin et al., 2011). Ultimately, provided the current practice parameters, and given that the collective effect size for CBT is greater than that of pharmacotherapy (1.45 versus 0.48, respectively; Watson & Rees, 2008), it appears that pharmacotherapy should be reserved for usage in combination with ERP for more severe and refractory cases (American Academy of Child and Adolescent Psychiatry, 2012).

Developmental Considerations The developmental stage of the affected child must be considered when implementing ERP for pediatric OCD. For instance, younger children often possess lower insight (Geller, Biederman, Faraone, Agranat, et al., 2001; Selles et al., 2014) and increased difficulty with implementing cognitive skills relative to their older counterparts (Kendall et al., 2006; Piacentini & Bergman,

2001), favoring primarily behavioral approaches in therapy. Additionally, psychotherapy for pediatric OCD typically entails a familial component, especially when children are younger (Freeman et al., 2012; Freeman et al., 2014; Lewin, Park, et al., 2014). Including family members in treatment holds the child accountable for completing out-of-session homework tasks, allows parents to model appropriate coping skills, and creates a cohesive family environment in uniting against OCD (Lewin & Piacentini, 2009; Piacentini, Langley, & Roblek, 2007). Family inclusion in therapy is particularly important when there is substantial symptom accommodation occurring.

Family accommodation of OCD symptoms includes modifications of family routines, facilitation of avoidance, and completion of rituals (Calvocoressi et al., 1995). While often completed with positive intentions (e.g., to reduce distress or mitigate time spent on compulsions), these maladaptive behaviors unfortunately serve to reinforce the negative cycle and maintain the symptomology (Calvocoressi et al., 1999; Storch, Björgvinsson, et al., 2010). With older youth, symptom accommodation can also extend to significant others and peers, requiring vigilance when addressing these behaviors in therapy. Collectively, family accommodation has been linked with various deleterious outcomes, including heightened functional impairment, OCD symptom severity, family conflict, as well as internalizing and externalizing behaviors (Caporino et al., 2012; Flessner et al., 2011; Peris et al., 2008; Storch, Geffken, Merlo, Jacob, et al., 2007; Wu, Lewin, Murphy, Geffken, & Storch, 2014). As such, prudent identification and cessation of family accommodation is imperative in disrupting the maintenance of OCD symptoms and increasing patient functioning (Thompson-Hollands, Edson, Tompson, & Comer, 2014).

Factors Affecting Treatment Response As many youth do not fully benefit from therapy for OCD, it is important to consider various factors that may impede the achievement of optimal treatment response. Various comorbidities may impact treatment response in youth, depending on the type of therapy employed. For instance, tics and externalizing behaviors may negatively impact response to pharmacotherapy (Ginsburg, Kingery, Drake, & Grados, 2008; March et al., 2007), while ADHD, DBD, and depression contribute to poorer response to CBT (Storch, Merlo, et al., 2008). Executive functioning may also impact treatment outcome, as the neuropsychological profile of OCD has illuminated difficulties with organization and recall of certain types of information (DuPaul, Eckert, & Vilardo, 2012; Greisberg & McKay, 2003). Indeed, this may impact their ability to independently and actively engage in various cognitive behavioral techniques (e.g., plan homework tasks and encode information learned from exposures), and poor decision-making is predictive of poorer pharmacological treatment response (Cavedini et al., 2002). Additionally, poor homework compliance in CBT has been linked to attenuated treatment response (Simpson et al., 2011; Simpson, Marcus, Zuckoff, Franklin, & Foa, 2012). Poor insight and inability to recognize the excessive nature of the OCD symptoms have also contributed to poorer response to treatment (Wu & Lewin, in press). Decreased treatment expectancies and previously failed trials of therapy are similarly related to lower treatment response (Lewin, Peris, Lindsey Bergman, McCracken, & Piacentini, 2011). Lastly, severe family accommodation and family conflict have also predicted poorer response to CBT (Ginsburg et al., 2008; Storch, Björgvinsson, et al., 2010). Given the variable treatment response of youth with OCD, it is important for clinicians to remain flexible in the implementation of treatment and tailor therapy to the idiosyncrasies of each family.

Treatment Adaptations Psychotherapeutic interventions for OCD are typically administered in an individual format, either with the child alone or with a family component (Freeman et al., 2014; Lewin, Park, et al., 2014; Piacentini et al., 2011; Thompson-Hollands et al., 2014). Exposure and response prevention can also be administered

in a group format, with group and individual modalities of ERP achieving similar treatment gains posttreatment (Barrett et al., 2004). Additionally, ERP can be implemented in an intensive format, allowing youth to complete therapy sessions multiple times a week versus the typical once-a-week protocol. Storch, Geffken, Merlo, Mann, et al. (2007) compared intensive and weekly modalities of ERP, demonstrating treatment gains in favor of intensive therapy following the acute phase of therapy. However, differences are dissipated by the follow-up assessments, indicating similar durability across both dosages of ERP. With the advent of technology, there has also been preliminary evidence supporting the use of telehealth mechanisms for delivering ERP to youth with OCD (Comer et al., 2014; Storch et al., 2011), demonstrating promise for the electronic dissemination of ERP to families in need.

There has been increased interest in the use of d-cycloserine (DCS), a partial agonist of the *N*-methyl-D-aspartate receptor. In an extension from animal studies, studies in humans have investigated the use of this antibiotic in potentially expediting the speed of obtaining treatment gains (Farrell et al., 2013; Kushner et al., 2007; Wilhelm et al., 2008). Studies have demonstrated mixed findings in pediatric OCD, with some studies finding no difference in treatment gains when compared to youth taking a placebo (Mataix-Cols et al., 2014), which was corroborated by a recent randomized-controlled trial (Storch et al., 2016); others found superiority of the DCS treatment group when looking at specific time points or different measures of OCD symptomology (Farrell et al., 2013). Storch, Murphy, et al. (2010) did not find a significant treatment group by time point interaction, but the effect size comparing the treatment groups at posttreatment favored DCS over the placebo. Collectively, DCS was initially thought to exhibit a mild-to-moderate effect on the augmentation of CBT for OCD (McGuire, Lewin, & Storch, 2014), but a recent comprehensive meta-analysis by McGuire et al. (2017) suggest that there may be no differences when compared to placebo augmentation.

Conclusion

Pediatric OCD is a heterogeneous psychiatric disorder that continues to affect a sizable amount of youth. Diagnostic criteria for OCD have remained largely unchanged since their inception (barring minor modifications in wording and specifiers), continuing to be characterized by intrusive obsessions and/or compulsions. Various factors complicate the clinical presentation and treatment of pediatric OCD, such as psychiatric comorbidities, developmental stages, and varying levels of insight, necessitating conscientious clinical judgment when working with this population. Looking toward the future, research trends are favoring the identification of neurobiological substrates and mechanisms underlying OCD (O'Neill et al., 2012), augmenting agents (e.g., DCS) used in conjunction with ERP, different dosages of therapy (e.g., stepped-care protocols, more intensive intervention), and third-wave therapies, such as acceptance and commitment therapy (ACT) and mindfulness-based CBT (Hopko, Lejuez, Ruggiero, & Eifert, 2003; Michael & Crowley, 2002; Ruiz, 2012; Twohig, Hayes, & Masuda, 2006; Van der Oord, Prins, Oosterlaan, & Emmelkamp, 2008). Given its association with various negative outcomes, more nuanced investigations into OCD, as well as prudent identification and timely treatment of this disorder, are crucial for the amelioration of OCD symptomology.

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Body Dysmorphic Disorder

11

Andrea S. Hartmann and Ulrike Buhlmann

Case

Tom is a 16-year-old boy who used to play soccer a lot, liked going to school to meet his friends, and regularly went to his guitar lessons. Upon presentation, he describes that for about 4 years he has continually decreased his activities and has even begun to hate going to school as he thinks that his face looks hideous. In particular, he is convinced that he looks stupid, believing that his eyes are too close together. Additionally, he hates his jaw line, which according to him is too round and not masculine enough. Every morning and evening, he spends about 2 h examining his face in the bathroom mirror. He tries to wear his hair over his face and uses a scarf to conceal his eyes and jaw line. On days when he feels especially bad about his appearance, he has begun to skip school, soccer practice, and guitar lessons. He explains that others might see him or he could see his own reflection somewhere. When prompted, he remembers two girls who seemed interested in him, which he could not understand, particularly as his frequent comparisons with his friends at school have led him to negatively evaluate his appearance. Even though he admits liking one of the girls, he insists that he would never go out on a date, fearing that the girl might see him close-up and be revolted. He has told his parents about his concerns, asking them for ideas on how to fix the problems. He describes that his parents have always told him that he looks just fine and that even his sister, who used to tease him at every opportunity, only commented that he was imagining it. When reporting the reactions of his family members, Tom gets agitated, as he cannot understand their misperception. He is convinced that his family and friends are merely trying to be nice when they say he looks fine, but are actually horrified by his appearance. He also fears that one day they will reject him because of his ugliness. His mother is terrified by the change in her son, who used to be a happy child who received compliments from everybody on his looks. She assumes that his striving for perfection in every area of his life (school, soccer, and guitar playing) has now moved on to his appearance. Tom also describes losing interest in everything apart from his appearance and tells that he will often sit in his room, feeling lonely, down, and doubting whether his life will get better in the future. Lately, he has begun to research surgical methods to change his jaw line and has run the findings by his parents in order to get their approval and financial support. His parents then made Tom's consultation appointment with a psychologist. Despite telling the psychologist about his problems, he makes it

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clear that he does not expect any help and does not understand why he has been sent to a psychologist instead of a surgeon.

Symptom Presentation

Tom is a fairly typical example of somebody with body dysmorphic disorder (BDD). The disorder is characterized by preoccupations with a perceived defect or flaw in one's own appearance, which might not be present or might only be slight according to others. However, for the individuals concerned, the "defects" often have a tremendous impact and they tend to describe them as hideous or ugly. Most individuals report concerns about their face or head (93%; Phillips, McElroy, Keck, Pope, & Hudson, 1993). Specifically, concerns often target the skin (in 80% of cases; being too wrinkled, having pimples or freckles or too many scars, or being too light in color), noses (39%; being hooked or too flat), lips (14.5%; being crooked, asymmetrical, too narrow, or too full), eyes (21.5%; having the wrong shape, being too narrow or too wide), or hair (57.5%; being too thin or thinning, the latter mostly in men). Other areas of concern include the shape of the face, the chin, teeth, ears, cheeks, forehead, facial muscles, overall body build, stomach, legs, breasts, genitals, buttocks, and arms (Phillips et al., 1993; Phillips, Menard, Fay, & Weisberg, 2005). The foci of concerns mentioned most often in children and adolescents are the skin, hair, stomach, weight, legs, and teeth (Albertini & Phillips, 1999; Phillips, Didie, et al., 2006). For most sufferers, the number of concerns about body parts increases and decreases (e.g., Phillips, 2005a), with an average of about six to seven areas. Phillips, Didie and colleagues (2006) showed that the frequencies of occurrence of the five most common concerns did not differ between adolescents and adults. The concerns are often obsessive in nature and are thus reported to consume a significant amount of time and to be difficult to resist and control. Furthermore, some individuals with BDD show poor or no insight, meaning that they have trouble realizing that they actually look normal. Some individuals also show delusions of reference (i.e., they are convinced that other people are talking about them or mocking them) (e.g., Phillips, 2004). It has been shown that the delusional and nondelusional variants of BDD do not differ on most demographics and clinical variables such as age of onset, course of illness, or phenomenology. However, individuals with delusional BDD often show more severe symptoms of BDD and delusions of reference (Mancuso, Knoesen, & Castle, 2010; Phillips, McElroy, Keck, Hudson, & Pope, 1994; Phillips, 2004). In addition, the level of insight often varies within an individual over the course of the disorder (Phillips, 2004). There are indications that BDD seems to be associated with more delusional beliefs in adolescents compared to adults (Phillips, Didie et al., 2006).

Most individuals with BDD engage in compulsive behaviors to check on, hide, or try to improve their perceived flaw (Phillips et al., 1993; Phillips, Menard, Fay, & Pagano, 2005). Ritualistic behaviors include camouflaging, excessive hair combing, makeup application, skin picking, or repeatedly asking others for reassurance. Further, mirror checking is very common (i.e., repeatedly checking the perceived flaw in every reflective surface). Some individuals spend hours in front of the mirror in the morning before they feel able to leave for work or school. Besides actual acts, individuals with BDD also engage in mental acts, e.g., comparing one's area of concern with the same area in others. Children and adolescents also exhibit these excessive repetitive or safety behaviors, most often camouflaging, asking for reassurance, comparing with others, and mirror checking (Albertini & Phillips, 1999), with no difference in their frequency to adults (Phillips, Didie et al., 2006). While all of these behaviors help to maintain the BDD symptoms in the long run (see etiology), some might even worsen the condition, e.g., when patients injure themselves while attempting to fix the perceived flaw (Veale et al., 1996). For example, Horowitz and colleagues describe a 16-year-old female adolescent who bound her breasts, eventually causing breast deformity (Horowitz, Gorfinkle, Lewis, & Philipps, 2002).

Most individuals with BDD avoid situations in which others might become increasingly aware of the flaw(s). For example, some might avoid daily activities (like school) or situations like

going to the beach or playground. Some might also try to avoid reflective surfaces to avoid an exacerbation of negative affect. In adult populations with BDD, avoidance can become so severe that sufferers might end up housebound (Phillips, Menard, Fay, & Pagano, 2005).

Some individuals' concerns focus distinctly on not being sufficiently large or muscular (Pope, Gruber, Choi, Olivardia, & Phillips, 1997). This BDD subtype, called muscle dysmorphia, mainly affects men and might be of importance in teenage boys. Core symptoms of muscle dysmorphia include compulsive and excessive weightlifting, rigid adherence to workout and dietary schedules, the use of performance-enhancing substances (e.g., anabolic steroids), body and/or mirror checking, and avoidance of situations in which one's body is exposed, or endurance of these situations with great distress (Olivardia, 2007). In most individuals with muscle dysmorphia, other BDD-related preoccupations are present as well. In general, individuals with do not differ from those BDD sufferers without muscle dysmorphia in demographic features, symptom severity, and delusionality. Pope and colleagues (2005) reported, however, that they are more likely to attempt suicide and have a poorer quality of life and a higher frequency of substance abuse – especially anabolic steroid abuse as described above.

In sum, research suggests that clinical features of BDD in adolescents and adults appear to be largely similar across age groups (e.g., Albertini & Phillips, 1999; Braddock, 1982; Cotterill 1981; El-Khatib & Dickey, 1995; Hay, 1970; Heimann, 1997; Phillips et al., 1994, 2006; Sondheimer, 1988), while no comparisons have been made with children.

History of the Diagnosis of Body Dysmorphic Disorder in the *Diagnostic and Statistical Manual of Mental Disorders*

BDD's unique symptomatology has been described in the literature for over 100 years. For example, Phillips (1991) describes the tradition of BDD in the European, Japanese, and Russian literature, most commonly referred to as dysmorphophobia (Morselli, 1891). Kraepelin (1909) and Janet (1903) both described the dysmorphophobic syndrome, classifying it as a compulsive neurosis and as a class of syndrome similar to obsessive-compulsive spectrum disorders, respectively. Many more authors have described the condition using different labels. However, only in the third version of the *Diagnostic and Statistical Manual of Mental Disorders (DSM-III)* was it mentioned for the first time, as an example of an atypical somatoform disorder, without diagnostic criteria or a delusional subtype (American Psychiatric Association (APA), 1980). BDD first became a separate disorder in the DSM-III-R, in the somatoform disorders section (APA, 1987), which included its new name as it was argued that the disorder does not involve phobic avoidance (Schachter, 1971). In the DSM-III-R, the delusional variant was also subtyped as a delusional disorder of the somatic type. In the DSM-IV, neither the criteria nor the allocation of the disorder was changed (APA, 1994). In the transition from the DSM-IV to the DSM-5 (APA, 2013), BDD was moved from the somatoform disorders section to the newly composed obsessive-compulsive (OC)-related disorders section. This section also includes obsessive-compulsive disorder (OCD), hoarding disorder, trichotillomania, and excoriation disorder. This move has been justified by various studies corroborating evidence of similarities of BDD with OCD in terms of its clinical presentation (for an overview, see Phillips et al., 2007). With the move, not only have the location of the disorder within the sections and the diagnostic system changed, but slight changes have also been made to the diagnostic criteria (see below). At no time in the history of the diagnosis, however, have child-specific criteria or age specifiers existed.

Current DSM-5 Diagnostic Criteria

In the move from the DSM-IV to the DSM-5, the wording of criterion A was changed from "imagined" to "perceived" flaw to make it more acceptable to individuals with BDD. Furthermore, criterion B was added to include compulsive

behaviors and mental acts. To fulfill the criterion, the individual needs to have performed repetitive behaviors (e.g., mirror checking or excessive grooming) or mental acts (e.g., comparing oneself with others regarding appearance) in response to his or her appearance concerns at some point during the course of the disorder. Criterion C excludes a diagnosis of BDD if it is not associated with significant distress and impairment. Finally, criterion D describes the differential diagnostic relationship with eating disorders, i.e., the concerns should not be better explained by an eating disorder. It is important, though, to consider that the two disorders can occur simultaneously and thus be diagnosed comorbidly. Furthermore, two specifiers were added. One describes the presence of MD, thus the dominance of the concern about low muscularity as well as the associated behaviors described above. The other describes the level of insight into the reality of the appearance beliefs, ranging from good to no insight. Given that the few studies which have examined BDD in children and adolescents found no major differences in BDD symptom presentation between younger age groups and adults, no age-specific criteria or specifiers have been included in the DSM-5 so far (APA, 2013).

Going back to 16-year-old Tom, we can establish that he suffers from concerns about perceived flaws that are not noticeable to others (criterion A). Additionally, he shows diverse behaviors in order to improve his appearance or camouflage it as well as mental acts in response to the concerns, i.e., comparing his face to those of his friends (criterion B). Furthermore, based on Tom's report, he is distressed by the concerns to the degree that he shows depressive symptoms, withdraws, and looks for drastic measures to fix the perceived flaws (criterion C). Finally, Tom does not show or report any signs or symptoms of an eating disorder (criterion D). Therefore, Tom should be diagnosed with BDD. In addition, with regard to the insight specifier, Tom is completely convinced that his perceived flaws are real and that his family and friends who contradict him are lying. He does not understand why he has been sent to a psychologist when the problem, at least with his jaw line, could be fixed so easily by a surgeon. Thus, we would characterize Tom as having completely delusional appearance beliefs.

Epidemiology and Course

Three representative population-based studies reported prevalence rates of BDD according to DSM-IV criteria in the general population. Specifically, Koran et al. (2008) reported a prevalence rate of 2.4% in the USA in adults aged 18 years and older, and Rief et al. (2006) and Buhlmann et al. (2010) described rates of 1.8% and 1.7%, respectively, in adolescents and adults aged 14 years and older in Germany. In all studies, women had slightly higher prevalence rates than men. Furthermore, data from Koran et al. (2008) suggest higher point prevalence rates in the younger subgroup (18–24 years, 4.4%) compared to older subgroups (25–34 years, 4.2%; 35–44 years, 3.7%; 45–54 years, 1.4%).

In addition, Mayville et al. (1999) examined self-reported BDD prevalence rates in high school students and obtained a fairly similar point prevalence rate of 2.2%, relative to the representative studies described above. Similar to findings derived from adult psychiatric inpatient samples (e.g., Grant, Kim, & Crow, 2001), prevalence rates in adolescent psychiatric inpatients ranged from 6.7% to 14.3% (Dyl, Kittler, Phillips, & Hunt, 2006; Grant et al., 2001). Moreover, higher prevalence rates have also been reported in dermatological treatment (7% and 15%; Phillips, Dufresne, Wilkel, & Vittorio, 2000; Sarwer, Wadden, Pertschuk, & Whitaker, 1998; Sarwer & Crerand, 2008) and reconstructive/plastic surgery settings (8.5%–15%; Sarwer & Crerand, 2008).

Age of onset of BDD is around 16 years (Bjornsson, Didie, Grant, Menard, Stalker, & Philips, 2013); however, patients are often not diagnosed until up to 15 years later (Phillips & Diaz, 1997; Veale et al., 1996). Probably, symptoms are often interpreted as age-appropriate developmental concerns, with non-pathological valence during teenage years. In a sample of adolescents with BDD, a significantly earlier age of

onset (M = 13.5, SD = 2.9) was reported (Philips et al., 2006); although this was a somewhat expected result given the fact that adolescents were examined, the authors suggest a cohort effect leading to a shift in age of onset due to increasing societal pressures with regard to appearance. There are also reports of cases with an early onset of BDD, at an age of as young as 6 years (Albertini & Phillips, 1999).

BDD shows a mean duration of 18 years and is a chronic disorder if left untreated (Phillips, Menard, Pagano, Fay, & Stout, 2006; Phillips, Pagano, Menard, & Stout, 2006). In a 1-year prospective study which also included individuals who were in treatment (84.2%), the probability of full remission was only 9%, and the probability of partial remission was 21% (Phillips, Pagano, et al., 2006). A more recent 4-year observational study (Phillips, Menard, Quinn, Didie, & Stout, 2013), in which 88% of participants received treatment during the observation period, reported a cumulative probability of 20% for full remission and 55% for full or partial remission. Unfortunately, no specific data are available on the course of BDD in children and adolescents.

Associated Impairments, Comorbidities, and Developmental Challenges

Individuals with BDD usually report a great impact on their psychosocial functioning. They often avoid social contacts and situations in which others might become aware of their perceived flaws (e.g., shopping trips, swimming, dating, and sexual relationships). Moreover, many individuals with BDD attend school or work only irregularly due to their appearance concerns (Didie et al., 2008; Phillips et al., 1993), and about 30% of BDD sufferers are even housebound. Additionally, individuals with BDD tend to be less frequently married and have a lower income (e.g., Rief et al., 2006). In children and adolescents, case reports have also illustrated that BDD is associated with poor grades at school (Phillips et al., 1995), stopping sports and other leisure activities (El-Khathib & Dickey, 1995), excessive school absences (Albertini et al., 1996), quitting high school (Phillips et al., 1995), social withdrawal (El-Khathib & Dickey, 1995), and being housebound (Albertini & Phillips, 1999).

Furthermore, individuals with BDD usually show poor self-esteem (e.g., Phillips, Pinto et al., 2004; Buhlmann, Teachman, Naumann, Fehlinger, & Rief, 2009; Hartmann, Thomas, Greenberg, Matheny, & Wilhelm, 2014) and poor quality of life (IsHak et al., 2012). In addition, morbidity is significantly higher in individuals with BDD compared to individuals with other mental disorders (Phillips & Menard, 2006). Specifically, BDD is associated with high levels of suicidal ideation (31–58% of all cases) as well as suicide attempts in 17–22% of sufferers (Buhlmann et al., 2010; Phillips et al., 1993; Phillips & Menard, 2006). In a prospective 1-year study, 3% of a clinical BDD group attempted suicide and 0.3% even committed suicide (Phillips & Menard, 2006). In adolescents, even higher lifetime rates of suicide attempts have been reported (80.6% a history of suicidal ideation and 44.4% attempted suicide (Phillips, Didie, et al., 2006)).

Comorbidity rates in BDD are high, with nearly all individuals suffering from at least one other mental disorder, often even three or more (Phillips et al., 1993). On average, individuals with BDD have two lifetime comorbid axis I disorders (Gunstad & Phillips, 2003). The most common comorbid diagnoses in order of occurrence are depression, social anxiety disorder (SAD), OCD, substance abuse disorder, and eating disorders (e.g., Gunstad & Phillips, 2003; Ruffolo, Phillips, Menard, Fay, & Weisberg, 2006). In general, the more comorbid disorders an individual is suffering from, the greater the functional impairment and morbidity they report (Gunstad & Phillips, 2003). Similar to the findings in adults with BDD, Albertini and Phillips (1999) showed in a BDD child and adolescent sample that the three most common comorbid disorders are depression, OCD, and SAD. The onset of SAD preceded the onset of BDD in 80%, in OCD only in 40%, and in depressive disorder only in 17% of all cases. Another study by Phillips

and colleagues also reported a high number of cases of substance abuse in adolescents with BDD and confirmed comparable comorbid profiles in adolescents and adults with the exception of panic disorder, which was significantly more common in adults (Phillips, Didie, Menard, Pagano, Fay, & Weisberg, 2006).

BDD in childhood and adolescence is associated with various developmental challenges. First, the developmental literature postulates a crucial role of body image influencing adolescent transitions (i.e., identity development, changing peer relationships, dating, and sexuality (Levine & Smolak, 2002)). Moreover, negative body image was shown to be associated with global self-esteem, depression, anxiety, and fear of negative evaluation by the peer group (Harter, Marold, & Whitesell, 1992; Levine & Smolak, 2002). These transitions, in turn, are also believed to have an important impact on the development of BDD, as adolescence is a period in which bodily changes are huge and appearance concerns are often prominent. As a consequence, it has even been suggested that BDD may be a pathological response to physical and physiological changes in this period (e.g., Andreasen & Bardach, 1977). Second, all of the associated impairments such as poor grades at school, stopping sports and other leisure activities, quitting school, and social withdrawal might adversely affect development, both socially and academically. Third, the high comorbidity rate of BDD further aggravates the poor developmental prognosis. Studies regarding depression, SAD, and OCD have shown these disorders' detrimental impact on development in this age group (Kovacs & Goldston, 1991; Ginsburg, La Greca, & Silverman, 1998; Piacentini, Bergman, Keller, & McCracken, 2003).

Differential Diagnoses

Differential diagnosis of BDD might be challenging. Particularly, it might be difficult to differentiate BDD from OCD, SAD, eating disorders, and depression (DSM-5; APA, 2013). However, the data gathered so far only stem from studies in adults. Table 11.1 provides an overview of similarities and differences between BDD and these disorders.

Etiology

There is no one single factor causing BDD, but the disorder is thought to be multifactorial: Biological, psychological, and sociocultural factors contribute to its development (Feusner, Neziroglu, Wilhelm, Mancusi, & Bohon, 2010).

As in many mental disorders, biological factors, i.e., genetic and neurobiological factors, are also likely to play a role in the development of BDD. So far, there is only one preliminary study indicating that the gamma-aminobutyric acid (GABA) $_{A}$-γ2 gene plays a role in the development of BDD (Richter et al., 2004). More specifically, the GABA $_{A}$-γ2 gene was found to have an association with BDD alone and BDD comorbid with OCD, but not with OCD alone. A recent study indicated that all OCDs share a genetic liability factor, while disorder-specific genetic factors were also apparent in OCD, BDD, and hoarding disorder (Monzani, Rijsdijk, Harris, & Mataix-Cols, et al., 2014). In terms of neurotransmitters, there is evidence of a serotonin (5-HT) dysfunction as well as irregularities in dopaminergic (DA) circuitry in individuals with BDD (Hadley, Newcorn, & Hollander, 2002). Furthermore, dysfunctions of the frontal striatal circuitry might also contribute to the development of BDD (Saxena & Feusner, 2006). Additionally, lower neural activity has been found in the visual association areas such as the parahippocampal gyrus, lingual gyrus, or precuneus when processing low spatial frequency pictures. Such pictures require a holistic rather than a detail-oriented processing of information. Thus, the finding underscores results from neuropsychological studies showing a bias toward detail focus (see psychological factors below) (Feusner, Hembacher, Moller, & Moody, 2011). No studies have been conducted specifically in children and adolescents to elucidate biological factors contributing to the development of the disorder. However, as BDD is often manifested in adolescence, Feusner

Table 11.1 Similarities with and differences from the most common differential diagnoses

Potential differential diagnosis	Similarities	Differences
Obsessive-compulsive disorder (OCD)	Intrusive thoughts Obsessions Repetitive behavior Age of onset, course of illness, sex ratio (Bienvenu et al., 2000)	Focus in BDD on appearance, in OCD on becoming ill, causing harm and other unwanted events BDD: less likely to be married, more likely to exhibit suicidal ideas and attempts, more substance dependence BDD: earlier onset of comorbid depression and SAD (Bienvenu et al., 2000) No focus on appearance- and social anxiety-related information in OCD (Buhlmann, McNally, Wilhelm, & Florin, 2002)
Social anxiety disorder (SAD)	Fear and avoidance of social situations Emotions: shame, fear of negative evaluations, and rejection Age of onset, sex ratio, course of the disorders (Fang & Hofmann, 2010)	Focus in BDD on appearance concerns, in SAD on possibly embarrassing, negatively evaluated behavior No compulsive behaviors in SAD (Fang & Hofmann, 2010)
Eating disorders (ED)	Disturbed body image and dissatisfaction (Hrabosky et al., 2009; Rosen & Ramirez, 1998) Repetitive behaviors (e.g., mirror checking, measuring of body parts, hiding body parts, over-exercising) (Hartmann, Greenberg, & Wilhelm, 2013)	In ED, body image concerns are only related to body shape and weight, and not to any other parts of the body as is the case in BDD Patients with anorexia nervosa (AN) deviate from normality in their weight, whereas patients with BDD have normal weight BDD shows more avoidance behaviors and greater delusionality than ED (Hartmann et al., 2015; Hartmann, Thomas, Wilson, & Wilhelm, 2013; Rosen & Ramirez, 1998) Sex ratio: in ED, the majority are girls and, in BDD, equal sex distribution (Buhlmann et al., 2010; Bulik et al., 2006; Koran et al., 2008)
Major depression (MD)	Poor self-esteem, rejection sensitivity, feelings of worthlessness (APA, 2013; Buhlmann et al., 2009; Park, Calogero, Young, & Diraddo, 2010; Pearson, Watkins, & Mullan, 2011; Sowislo & Orth, 2013)	No underlying obsessions and compulsions in MD Earlier age of onset and more chronic course in BDD (Richards, 2011) (Selective) serotonin reuptake inhibitors ([S]SRIs) for both, but BDD takes more time to respond and needs higher dosage (Perugi et al., 1996; Phillips & Hollander, 2008)

and colleagues suggested that hormonal and physical changes in appearance might be an eliciting factor (Feusner, Neziroglu, et al., 2010).

Psychological factors that contribute to BDD can be divided into personality, perceptive, cognitive, and emotional aspects. In terms of personality, individuals with BDD report high levels of perfectionism, which may lead to unrealistic standards, also in terms of attractiveness (Buhlmann, Etcoff, & Wilhelm, 2008; Hartmann, et al., 2014). Combined with an increased fear of disapproval (i.e., rejection sensitivity, Kelly, Didie, & Phillips, 2014), perfectionism might enable the development of BDD (Buhlmann et al., 2008). Further personality characteristics in individuals with BDD are a high level of neuroticism and a low level of extroversion (Phillips, Siniscalchi, & McElroy, 2004). Regarding facial perception, a misperception of one's own and others' attractiveness are found in BDD (Buhlmann et al., 2008). When combined with the maladaptive cognitive beliefs about attractiveness (i.e., the belief that one must look perfect to receive acceptance and love), this might evoke negative emotions such as shame and contribute to the development of BDD (Buhlmann et al., 2009; Buhlmann, Teachman, &

Kathmann, 2011). In line with the abovementioned neuroanatomical characteristics, neuropsychological studies have shown that individuals with BDD tend to focus on and remember details rather than the entire whole (Deckersbach et al., 2000). This detail bias has been reported not only for general objects (Feusner, Hembacher, Moller, & Moody, 2011) but also for faces (Feusner, Townsend, Bystritsky, & Bookheimer, 2007). This selective attention and processing of information, in particular when focusing on one's own body and its flaws, might foster the BDD symptomatology. With respect to emotional factors, difficulties in emotional expression recognition have been shown in BDD. In particular, emotionally neutral facial expressions are misinterpreted as contemptuous or angry when perceived as self-referential but not other-referential (Buhlmann, McNally, Etcoff, Tuschen-Caffier, & Wilhelm, 2004; Buhlmann, Etcoff, & Wilhelm, 2006). This difficulty might account in part for delusions of reference in BDD. Several other results have shown that there are difficulties in identifying emotional expressions overall, independent of the emotion shown (e.g., Feusner, Bystritsky, Hellemann, & Bookheimer, 2010). To date, no neuropsychological or experimental clinical psychological studies have confirmed these characteristics in children or adolescents.

Finally, when focusing on sociocultural factors, it becomes clear that today's society values appearance highly and considers it to be the key to success and acceptance among one's peer group. The media support this notion by portraying attractiveness as an indicator of intelligence and moral behavior. Furthermore, they show what beauty is supposed to look like (Neziroglu, Khemlani-Patel, & Veale, 2008). On a less societal and more individual level, a retrospective study showed that individuals with BDD report significantly more appearance- and competency-related teasing in childhood compared to a mentally healthy control group (Buhlmann, Cook, Fama, & Wilhelm, 2007). Others have described correlations between BDD and sexual, emotional, and physical abuse as well as neglect in childhood (Didie et al., 2006; Neziroglu, Khemlani-Patel, & Yaryura-Tobias, 2006).

An individualized model, as it could be put together for a patient like 16-year-old Tom, is depicted in Fig. 11.1. It is based on the scarce knowledge regarding etiological factors outlined above as well as the individual experiences of the patient.

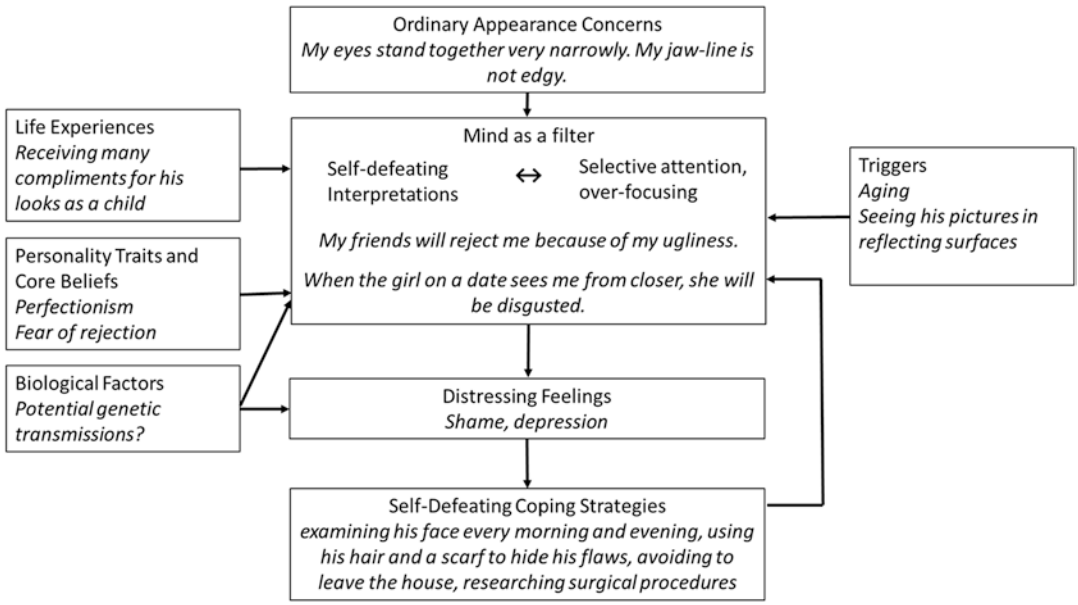

Fig. 11.1 Individualized etiological model for Tom (Adapted from Cognitive behavioral therapy for body dysmorphic disorder, Wilhelm, Phillips, & Steketee. 2013. Copyright Guilford Press (Reprinted with permission of The Guilford Press))

Diagnosis and Treatment

Despite the resulting distress and consequences in daily functioning, few adult individuals with BDD seek psychological or psychiatric treatment directly. Reasons for this reluctance are logistics and financial concerns, stigma, shame, and skepticism about treatment (Buhlmann, 2011; Marques, Weingarden, Leblanc, & Wilhelm, 2011). When individuals with BDD do seek treatment, in particular if they are highly delusional, it is frequently dermatological or surgical in nature, which is more in line with the wish to fix the perceived flaw (e.g., Phillips et al., 2000; Sarwer & Crerand, 2008). Sarwer and Crerand summarized retrospective studies which suggest that these treatments are rarely helpful for BDD symptoms (Sarwer & Crerand, 2008). Thus, more information about BDD for primary care physicians (such as in Slaughter & Sun, 1999; Phillips & Dufresne, 2002), plastic surgeons, and dermatologists (Phillips & Dufresne, 2000) is warranted to assist them in identifying the patients and to promote referrals to effective treatments. No data are available on the treatment-seeking status of children and adolescents with BDD. One might assume that the percentage of youth reaching psychological and psychiatric practices is even lower, given the lacking knowledge about the disorder and its potential attribution to developmentally specific and normative concerns.

Diagnosis

Upon referral to a specialist, individuals with potential BDD should be properly diagnosed. As a screening tool, the Body Dysmorphic Disorder Questionnaire (BDDQ; Phillips, Atala, & Pope, 1995) can be used to establish a self-report-based BDD diagnosis. This screening instrument has an adolescent version with good psychometric properties (Phillips, 2005a; Grant, Kim, Grow, 2001). If time permits, the employment of a structured clinical interview is essential. For adults and adolescents, the BDD module of the Structured Clinical Interview for DSM-IV (First, Spitzer, Gibbon, & Williams, 2002) can be used. Severity of BDD can be measured using the Yale-Brown Obsessive-Compulsive Scale adapted for BDD (BDD-YBOCS; Phillips et al., 1997) or the Body Dysmorphic Disorder Examination (BDDE; Rosen & Reiter, 1996) in adults and adolescents. In order to assess the level of delusionality, the Brown Assessment of Beliefs Scale (BABS; Eisen et al., 1998), a short expert interview, is useful. For children, neither structured interviews nor self-report instruments are available to assess BDD symptomatology. Thus, there is an immense need to develop such instruments for this age group.

Treatment of BDD

Empirical Support for Treatment The currently recommended treatments for BDD in adults are cognitive behavioral therapy (CBT), serotonin reuptake inhibitors (SRIs), or a combination of the two (e.g., Ipser, Sander, & Stein, 2009). In several studies, the SRIs clomipramine, fluvoxamine, fluoxetine, citalopram, and escitalopram have proven to be effective for 53–73% of patients (for an overview, see Phillips & Hollander, 2008). Notably, the dosage is usually higher than in depression, and the response is slower and more gradual and appears over several months, as has also been shown in individuals with OCD (Phillips & Hollander, 2008). Further studies also suggest that following trials with different SRIs at an optimal dose showing non-adequate responses, augmentation with neuroleptics might be considered, even though their effectiveness has only been demonstrated in a few studies (e.g., Phillips, 2005b, 2005c). In sum, there are promising indications for pharmacological treatment of BDD; however, adequate large-scale randomized controlled trials are still sparse. In addition, data on pharmacological treatment of BDD in children and adolescents are rare. A small number of case studies and case series support the use of SRIs (fluvoxamine, fluoxetine, paroxetine, and clomipramine) in this age group (Albertini et al., 1996; Albertini & Phillips, 1999; Horowitz et al., 2002; Phillips et al., 1995). Given the concerns about suicidality and SRI use in youth (Adegbite-Adeniyi, Gron, Rowles, Demeter, Findling, 2012), caution is warranted when prescribing them in this age group.

The evidence in terms of CBT for BDD in adults stems from four randomized controlled trials (RCTs). Two older RCTs reported favorable outcomes for both an individual (Veale et al., 1996) and a group CBT intervention (Rosen, Reiter, & Orosan, 1995). Two recent treatment studies provided support for the effectiveness of a modular CBT in an individual setting (Wilhelm et al., 2014) and a superiority of CBT compared to anxiety management in an individual setting (Veale et al., 2014). Furthermore, a recent feasibility trial showed encouraging effects of a 12-week internet treatment program. Such an implementation would lead to greater accessibility of treatment for this patient group (Enander et al., 2014). The findings from the RCTs have been further corroborated by results from open trials and case series (McKay, 1999; McKay et al., 1997; Wilhelm, Otto, Lohr, & Deckersbach, 1999). In children and adolescents, several case studies hint at the success of CBT in these age groups (Aldea, Storch, Geffken, & Murphy, 2009; Phillips et al., 1995; Horowitz et al., 2002; Sobanski & Schmidt, 2000). However, future research, in particular in the form of RCTs, is needed to further explore this, particularly in youth.

Description of CBT CBT for adults with BDD consists of psychoeducation, cognitive restructuring, behavioral experiments, exposure and response (ritual) prevention, perceptual retraining, and habit reversal. The components will be summarized in the following.

In psychoeducation, in a first step, the therapist provides general information about BDD, e.g., typical symptoms and prevalence rates. Then, components of CBT as well as evidence for the treatment are discussed. Patient and therapist continue by developing an individualized model of BDD based on the patient's background and symptoms as well as the etiological model of BDD (see etiology and, e.g., Wilhelm et al., 2013). From this individualized model, CBT techniques can be derived, and the model can be used for tailoring intervention strategies to the patient's specific needs later in treatment.

Cognitive restructuring is used to identify, evaluate, and subsequently modify negative thoughts and beliefs the patient has about his or her appearance. The patient further learns to evaluate whether or not the beliefs are realistic and helpful and, if not, to generate more accurate alternative thoughts (e.g., Wilhelm et al., 2013). The therapist can start by explaining thinking errors to the patient, e.g., "all or nothing thinking ("I am either beautiful or monstrous, hideous, and ugly"). While monitoring negative and automatic appearance-related thoughts, ideally with a thought record, the patient can learn to identify the thinking errors." Whenever the patient is distressed, he or she should try to capture the triggering event and negative automatic thought including the inherent thinking error. Additionally, patients are taught Socratic questioning for use in these distressing situations or situations that they are avoiding ("Why do I always feel so stressed out about my appearance when I go to a party with my boyfriend?", "Perhaps I am worried that no one will like me due to my bad skin?"). Once identification of thoughts has been established, the therapist assists the patient in evaluating the validity ("Is there any evidence that nobody likes me because of my bad skin?") or usefulness of the thought ("Does it help me to think I can only have a good time if I don't break out in pimples anymore?"). Together, the pros and cons of holding onto the appearance-related thoughts can be examined and weighed ("It is beneficial to hold onto the thought because it helps me feel in control, but on the other hand, I can't enjoy myself anymore when I go out.") (Wilhelm et al., 2013). Further cognitive techniques are the pie chart technique or role playing. In a next step, the therapist can assist the patient in exploring core beliefs relating to appearance concerns (e.g., "I am unlovable") using Socratic dialogue. Techniques used for exploration are the downward arrow technique, i.e., the therapist will keep asking the patient "What would it mean if you…," starting from the patient's automatic negative thought of "People will think I am ugly because of my pimples," usually until the core belief is reached ("If people notice my pimples, they won't like me, and this means that I am not lovable"). The core beliefs can then be addressed through cognitive restructuring as previously described or behavioral experiments in which core beliefs are tested for their validity in real life (Wilhelm et al., 2013).

The second core strategy of CBT is exposure and response prevention (e.g., Wilhelm et al., 2013). After therapist and patient have discussed the rationale behind exposure exercises, they develop a list of situations that evoke anxiety in the patient and are usually avoided. These situations are then arranged into a hierarchy with regard to the extent of avoidance and associated anxiety. Frequently reported situations include walking around in public without wearing specific camouflaging items of clothing, leaving the house without wearing makeup, or going to the beach. Exposure exercises begin with situations at the lower end of the hierarchy (low in anxiety and avoidance). Notably, it is important to stay in the situation for a sufficient amount of time for a decrease in anxiety to occur – usually, habituation sets in after an initial anxiety increase. The exposure exercises should be practiced repeatedly and frequently, continuously moving upward in the situation hierarchy. Ritual prevention, the accompanying strategy, focuses on the prevention of distraction and accordingly on allowing a greater experience of the anxiety and a more comprehensive habituation. In a first step, therefore, a patient's rituals and avoidance behaviors are assessed. Next, specific goals for eliminating these behaviors are set, albeit taking into account that a full elimination of all rituals at once might be difficult. Therapists might then suggest the following steps: delaying the response, including a continuously increasing delay period, or reducing aspects of rituals (e.g., putting on makeup only once rather than twice in the morning) (Wilhelm et al., 2013).

The third core strategy of CBT is perceptual retraining. This is included in the treatment plan if patients have established a dysfunctional behavior regarding mirrors such as avoiding them, checking them over and over again, or both. It is also a valuable technique if patients show selective attention toward particular body parts that they perceive as ugly. Such behaviors may maintain negative thoughts and feelings about appearance (see etiology). Mostly, perceptual retraining is conducted through mirror exposure. Of note, for mirror exposure, the mirror needs to be large enough to show the entire face and/or body and must be placed in a well-lit area, and the patient should be instructed to stand 1 foot away if looking at the face and 2.3 feet away if looking at the whole body. When starting the exercise, the patient is asked to examine the different body parts (head to toe) and to describe them in as much detail as possible in an objective, nonevaluative fashion. This detailed, neutral, and nonjudgmental description fosters a less distorted manner of perception of the patient's own body and a healthier relationship with the mirror. Here too, ritual prevention is essential, i.e., behaviors typically used in front of mirrors should be avoided.

Finally, habit reversal can be an additional strategy used in CBT, for compulsive behaviors such as skin picking, hair plucking, and body touching (Veale & Neziroglu, 2010; Wilhelm, et al., 2013). First, the patient describes the behavior in a diary in as much detail as possible in order to increase awareness of the behavior (length and situations in which it occurs). Then, a competing response is introduced; thus, another behavior is performed that is incompatible with the original behavior.

Potential Difficulties in Treatment Difficulties once the treatment has started can include low motivation, ambivalence about the treatment, comorbid depression, and suicidality. Lack of motivation and ambivalence at the start of treatment in BDD can often be due to poor insight and the belief that cosmetic procedures would be the better treatment option. Motivational interviewing (MI), which has been adapted by Wilhelm et al. (2013) for use in BDD, can be useful in this case. Specific aspects include the focus on the distress caused by the thought (Let's try to reduce the distress"). Furthermore, nonjudgmental Socratic questioning of pros and cons of the CBT treatment or of the discrepancy between BDD symptoms and the patient's goals ("What should your life look like 10 years from now?") can also be helpful. Additionally, a focus on the usefulness rather than the validity of the belief can be constructive, particularly in highly delusional patients (e.g., "Are your beliefs preventing you from participating in activities you enjoy?"). Later on in

the treatment, MI strategies might help to combat ambivalence about specific treatment strategies (e.g., exposure exercises).

Comorbid depression can interfere with treatment due to lack of energy or concentration difficulties. Therefore, it might be helpful to target depressive symptoms first or in parallel through adjunctive pharmacological therapy. Regular monitoring of the patient's suicidal ideation, intent, and plans is essential, and if there is acute suicidality, a higher level of care may become necessary, e.g., full or partial hospitalization (Wilhelm et al., 2013).

Considerations for Treatment in Adolescents Only a small number of case studies have investigated the effectiveness of CBT in youth (Aldea et al., 2009; Greenberg et al., 2010, Horowitz et al., 2002; Sobanski & Schmidt, 2000). As only one report is available for children (Albertini & Phillips, 1999), we will focus on adolescents. Greenberg et al. (2010) point out that CBT for adolescents differs from that for adults in that it must take into account age and developmentally appropriate tasks and transitions and the involvement of the parents. For example, some researchers had parents assist in the treatment of their children. Therapists might provide psychoeducation about BDD to parents, advise them to reduce accommodation of rituals (e.g., reassurance, not to buy beauty products), and have them help to maintain adolescents' motivation for treatment. Other treatments focused directly on the whole family, i.e., family-based CBT (Aldea et al., 2009). The inclusion of parents in treatment makes it essential to clarify issues of confidentiality at the beginning of the treatment when working with youth, enabling them to have a say in what will be told to their parents. Adolescents' motivation for treatment may be particularly low, in particular as they are usually brought in for consultation by their parents. Motivation might be further lowered by low insight typical for youth and a potential lack of metacognitive abilities. Therefore, the above-mentioned MI strategies might be particularly useful in this age group. Furthermore, besides BDD-specific treatment components, adolescent transitions and tasks need to be addressed, i.e., autonomy from family, and peer or romantic affiliations, as they may interact with BDD (Greenberg et al., 2010; Phillips & Rogers, 2011).

Summary and Outlook

BDD is a debilitating and impairing mental disorder that has its onset in adolescence, with a few mentions of childhood cases. Clinical presentation in youth and adults looks fairly similar. However, despite the relevance of body image and the potential damage on, for instance, the development of identity by such a severe body image disturbance, research on BDD in youth is rare. Factors identified as maintaining factors of the disorder in adults have not been confirmed in youth, and successful treatment components in adults (CBT and SRIs) have also not yet been tested in youth in large randomized controlled trials, but have been tested in case studies and case series only.

Thus, there is pressing need to develop evidence-based treatments for youth along the lines of CBT, which has proven successful for adults with BDD. Furthermore, studies on the etiology of BDD should also be broadened to this age group in order to gain more understanding of age-specific aspects of the disorder. Finally, information for colleagues from other disciplines such as cosmetic surgeons and dermatologists, but also primary care physicians and pediatricians, is necessary. This should ensure that the identification of BDD, which can be easily mistaken for normative developmental concerns or other mental disorders in this young age group, is enhanced and referrals to specialists are made.

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Trichotillomania

12

Martin E. Franklin and Sarah Morris

Trichotillomania: History in DSM

TTM has been described in the medical and psychological literature for many decades (e.g., Hallopeau, 1889) and was originally included in the DSM-III-R in 1987 (American Psychiatric Association, 1987) along with Impulse Control Disorders Not Elsewhere Classified due to the commonly reported failure of TTM sufferers to refrain from urges to pull out their own hair. However, this grouping associated TTM with disorders that might be considered strange bedfellows phenomenologically and clinically (e.g., kleptomania, intermittent explosive disorder, pathological gambling). Indeed, the inclusion of TTM among a diverse group of impulse control problems such as pyromania was openly questioned (e.g., McElroy et al., 1992), with some researchers theorizing that TTM was better described as a "nervous habit" with body foci similar to face and other skin picking, bruxism, and nail biting (Christenson & Mansueto, 1999) or, perhaps more broadly, as a body-focused repetitive behavior (BFRB; Zohar & Arush, 2012).

Despite this ongoing debate in the literature, TTM continued to be housed in DSM-IV TR (American Psychiatric Association [APA] 2000) with the impulse control disorders and defined by hair pulling that results in noticeable hair loss, increasing tension immediately prior to pulling, gratification or relief when engaging in pulling, and significant distress/functional impairment. In addition to debate about the proper placement of TTM within the DSM classification scheme, adult and pediatric studies raised specific questions about the appropriateness of TTM Criteria B (mounting tension prior to pulling) and C (gratification or relief following pulling) as requirements for diagnosis. Notably, 17% of Christenson et al.'s (1991) sample of adult treatment-seeking chronic hair pullers failed to meet these criteria; similar findings were reported more recently from a South African adult sample (du Toit et al., 2001). Interestingly, these two phenomena are positively related – people who experience tension prior to pulling also tend to experience relief after pulling (du Toit et al., 2001). Problems applying these criteria may be especially pronounced in young children, who typically find it challenging to describe their affective states (Hanna, 1997; Reeve, 1999). In our clinical sample of 48 children and adolescents ages 7–17 inclusive evaluated for participation in TTM treatment studies, 81% endorsed Criterion B and

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83% endorsed Criterion C (Tolin et al., 2007). More recent studies have also raised questions about the incremental and predictive validity of Criteria B and C (e.g., Conelea et al., 2012; Houghton et al., 2015; Lochner et al., 2011; Stein et al., 2010), to the point where they were the primary focus of debate on the DSM subcommittees and workgroups tasked with making final recommendations for the DSM-V document.

The scientific evidence and collective clinical wisdom up to that point indicated that DSM-IV Criteria B and C may not apply to all those who pull hair to a clinical degree, and the criteria may not be developmentally sensitive. Thus, strict adherence to these criteria could result in exclusion of patients who meet the hair pulling and functional impairment criteria yet are unable to articulate their affective states before and after pulling in a manner fully consistent with Criteria B and C. Multiple studies attesting to a lack of important clinical differences between patients who met or did not meet Criteria B and C versus those who did not served as the final straw upon the camel's back. Accordingly, this issue was finally remediated by the DSM-V subcommittee on TTM, whose proposal to eliminate both as formal requirements for a diagnosis of TTM was upheld for the new DSM-V manual.

DSM-V Criteria for Trichotillomania

The revised criteria for TTM in DSM V are as follows:

A. Recurrent pulling out of one's own hair, resulting in hair loss
B. Repeated attempts to decrease or stop hair pulling
C. Clinically significant distress or impairment in functioning
D. Not attributable to a general medical condition (e.g., alopecia)
E. Not better explained by another mental disorder (e.g., body dysmorphic disorder)

The removal of DSM-IV-TR Criteria B and C does not eliminate their relevance to the practicing clinician: it should be noted that the research studies cited above indicated that most patients, especially adults, still endorsed these phenomenological experiences. Rather, the intent of removing them was to eliminate the *requirement* that they be present in order to make a TTM diagnosis. This more flexible diagnostic system will likely allow clinicians to use the TTM diagnosis in cases where pulling and distress/functional impairment are present and not better accounted for by other conditions; it was believed that this would permit more frequent use of the diagnostic entity for children in particular, for whom reporting on psychological antecedents and consequences of their behavior poses a daunting developmental challenge for some. Researchers who study TTM will likely continue to ask about and record information about premonitory urges and affective experiences associated with pulling, as examining whether they are associated with certain styles of pulling (e.g., focused pulling in response to clear affective cues vs. automatic pulling that takes place outside awareness – see Flessner et al., 2007, 2008) and with treatment response.

Etiology

Theories of TTM's etiology abound (e.g., Grant et al., 2007), but as yet there is insufficient data available as yet to clearly identify or isolate a single cause of the disorder. Recent studies have identified a genetic component (Novak et al., 2009), in that greater TTM diagnostic concordance was observed in monozygotic as opposed to dizygotic twins; a family proband study was convergent with these findings in that there was greater risk for TTM and OCD in the family members of TTM cases compared with controls (Keuthen et al., 2014). A comprehensive review of neurobiological theories of TTM is beyond the scope of our paper (see Chamberlain et al., 2007, for such a review), but it also must be stated that learning theory has been brought to bear to explain why individuals begin pulling their hair; these neurobiological and behavioral accounts should not be considered mutually exclusive. The factors

associated with TTM onset are likely complex and intertwined; moreover, these etiological factors may or may not be the same factors associated with maintenance. Most behavioral or cognitive-behavioral accounts of TTM focus on the factors that maintain pulling rather than those that cause it, which allows clinicians to focus on the immediate antecedents and consequences of pulling to identify points for behavioral intervention (for a comprehensive review, see Franklin & Tolin, 2007). In particular, positive and negative reinforcement cycles are posited to maintain TTM behavior: for example, if an individual responds to an aversive physiological sensation such as a premonitory urge by pulling out a hair and the process of pulling out the hair yields relief from the aversive experience or produces a pleasurable sensation, then that cycle will be strengthened, resulting in more pulling down the line. Clinically with TTM it is possible to see either of those patterns (positive or negative reinforcement cycles) and sometimes even both patterns evident in the same person depending on other environmental factors (e.g., degree to which the person's attention is drawn to another task such as reading or watching TV).

Symptom Presentation

Although comprehensive, large-scale epidemiological, and longitudinal studies have yet to be conducted, TTM is estimated from smaller studies to affect 1–3.5% of late adolescents and young adults (e.g., Christenson et al., 1991); rates among younger children unfortunately remain unknown (Tolin et al., 2007). Across the developmental spectrum, sufferers can experience medical complications such as skin irritations at the pulling site, infections, and repetitive-use hand injuries (du Toit et al., 2001). The subset of individuals with TTM who ingest the hairs after pulling, which has been estimated to be as high as 20% (Grant & Odlaug, 2008), are at risk for gastrointestinal complications stemming from trichobezoars (i.e., hairballs; Bouwer & Stein, 1998; Grant & Odlaug, 2008), which have been documented in children as young as four (Lanoue & Arkovitz, 2003). Notably, TTM onset in childhood or adolescence appears to be the norm and appears to precede that of most comorbidities (Christenson & Mackenzie, 1995).

Less information is available on TTM presentation in youth, but the available literature is convergent with information about clinical features of adult hair pulling. As with adults, the scalp is the most common pulling site in children and adolescents, followed by eyelashes and eyebrows (Franklin et al., 2008; Reeve, 1999; Tolin et al., 2007). Notably, our pediatric TTM open clinical trial's rate of 27% male participants is considerably higher than is typically reported in clinical studies of TTM in adults (Tolin et al., 2007); this roughly 3:1 male-to-female ratio more closely resembles what has been found in college survey studies of hair pulling, including our own (Hajcak et al., 2006). This disparity may have to do with treatment-seeking behavior: perhaps because *parents* are inquiring about services for children and adolescents; this factor offsets the tendency evident in adult men to avoid psychotherapy in general and to avoid services for TTM specifically.

With respect to the pulling process itself, in our clinical work, we have seen that the most common method of pulling involves isolating a specific hair with the thumb and index finger, pulling that hair and dropping it, and then returning almost immediately to pull another one in the same manner. Although such a pattern may reflect the modal patient's pulling behavior, a subset will inspect the hair after removing it from the pulling site: some will then visually examine or feel the root if they were able to harvest one, and a subset of those patients play with the hair, roll it between their fingers, touch it to their face or lips, or insert the root or whole hair into their mouths. Grant and Odlaug's study (2008) indicated that about 20% of hair pulling adults ate part of the hair or the whole hair – such data have not been collected formally in youth as yet, but most certainly this possibility should be evaluated with any patient who engages in pulling behavior.

Associated Impairment

The effect of TTM on functional outcomes has been a topic of increased study over the past decade (e.g., Franklin et al., 2008; Woods et al., 2006), and what has become evident from these and other investigations is that TTM is far from trivial in terms of its impact. Adults with TTM report impaired school, work, and social functioning, lowered career aspirations, and missed work days (Diefenbach et al., 2005; Seedat & Stein, 1998; Woods et al., 2006). Adults also report spending considerable financial resources on concealment methods and on treatments with varying degrees of success (Wetterneck et al., 2006). Because TTM usually strikes during sensitive developmental years, it can be especially disabling (Rothbaum & Ninan, 1994); indeed, TTM has been found to be at least moderately impairing in the social and academic realms for older children and adolescents (Franklin et al., 2008; Tolin et al., 2007; Panza et al., 2013). Many adolescents with TTM encountered in our clinic express trepidation about the possibility that their classmates and friends will discover their bald patches and evaluate them negatively as a result. As it turns out, unfortunately, such concerns about peer rejection may be well founded: developmentally normal eight graders viewing videotaped segments of actors portraying individuals with TTM, chronic tic disorders (CTDs), or neither condition rated the social acceptability of those with TTM and CTDs as significantly lower than those without either condition (Boudjouk et al., 2000). Pulling can also negatively impact family functioning, contributing to family arguments and secrecy, which in turn can increase stress and exacerbate TTM symptoms (Moore et al., 2009; Stemberger et al., 2000). A more recent study examining family environment in adolescents with TTM, their parents, and a matched adolescent control group indicated that youth with TTM reported more expression of anger, aggression, and conflict in their families compared to that observed in controls; moreover, there was a significant discordance between adolescents with TTM and their parents with regard to their perceptions of the family environment (Keuthen et al., 2013). It is unclear, however, whether these family difficulties are causal or largely consequent to the development of TTM; longitudinal research is sorely needed to address this important question.

Comorbidity

Psychiatric comorbidity in adults appears to be very common, with anxiety disorders, mood disorders, substance use disorders, eating disorders (Christenson et al., 1991; Woods et al., 2006), and personality disorders being the most common comorbid conditions in adults (Christenson et al., 1992), and anxiety and disruptive behavior disorders are commonly observed in youth (Christenson et al., 1991; King et al., 1995; Panza et al., 2013; Tolin et al., 2007).

In our descriptive study in youth with TTM (Tolin et al., 2007), we found that the typical participant had clearly visible bald patches or thinning as measured by IE ratings of alopecia and reported on average 30–60 min per day of hair pulling. With respect to academic functioning, 79% of parents reported that their child had academic problems, including 44% reporting that their child had difficulty completing classwork and homework; the specific relationship between time lost to pulling and concealment efforts and academic functioning has yet to be explored (Tolin et al., 2007). Social functioning can also be impacted: children and adolescents, already sensitive about their appearance, often go to great lengths to hide pulling sites and avoid activities that might lead to discovery of the problem by their peers (e.g., swimming, sleepover parties, etc.). Avoidance often results in receiving fewer subsequent invitations to participate in activities, compounding their sense of isolation. When pulling is discovered by peers, youngsters often experience a great sense of shame and embarrassment and can experience teasing and peer rejection. Pulling can also negatively impact family functioning, contributing to family arguments and secrecy (Stemberger et al., 2000). It is unclear whether these difficulties are causal or consequent to the development of TTM; longitudinal research is sorely needed to address this important question.

Course

One of the most vexing problems confronting the field is the lack of longitudinal data regarding the course of illness, as it is unclear from the literature at present what percentage of affected individuals will continue to have symptoms over time, whose symptoms will remit entirely, whose symptoms will wax and wane, and what clinical, biological, and demographic factors predict long-term symptom status. This may be especially important in the case of very young children (ages 18 months–5 years), since there is no guidance regarding whether parents should simply wait until the child is older and sufficiently self-aware to recognize urges and intervene using cognitive-behavioral interventions. Clinical experts posited at one time that pulling in very young children represented a more benign form of the disorder (Swedo & Leonard, 1992); however, a subsequent case series of children ages 5 and younger indicated that anxiety disorders and high levels of family distress were evident (Wright & Holmes, 2003), which may suggest otherwise. Walther et al. (2014) attempted to characterize hair pulling in young children in a web-based survey of parents whose children engaged in hair pulling behavior, and their findings indicated that preschool-aged youth (5 and younger) had comparable parent-reported pulling frequency rates compared to older children (ages 6–10), but that the older group was more aware, more impaired, and had higher rates of comorbid psychopathology. The absence of a group of young pullers followed over time limits the utility of these comparisons in establishing a clear developmental trajectory for TTM, but the findings do suggest that TTM's impact and complexity may well worsen over time.

Developmental Challenges

A major priority in TTM psychopathology and treatment research is to recruit younger samples, with the goal of improving our understanding of TTM closer in time to its onset and, by extension, treating TTM effectively earlier. Treating TTM earlier will perhaps reduce future functional impairment and prevent the development of debilitating comorbid disorders. The few studies that have examined TTM and its treatment in younger samples document the presence of TTM in youth ranging from toddlers to adolescents (Franklin et al., 2008; Walther et al., 2014) and have suggested its responsiveness to behavioral interventions even at younger ages (Franklin et al., 2010, 2011). Nevertheless, despite the fact that TTM appears to be a relatively common pediatric onset disorder associated with significant morbidity, comorbidity, and functional impairment in adults (Woods et al., 2006), surprisingly few TTM psychopathology research studies have actually included adolescents or children. There is only one published randomized controlled trials (RCTs) of a psychopharmacological interventions for youth with TTM (Bloch et al., 2013 – see below), and that trial failed to support the efficacy of NAC in youth despite a positive trial in adults (Grant, Odlaug, & Kim, 2009). Although the initial findings for cognitive-behavioral therapy for pediatric TTM have been encouraging (Franklin et al., 2010, 2011 – see below), key questions remain regarding the role of developmental factors in TTM psychopathology and treatment response. With respect to similarities and differences in TTM phenomenology across the developmental spectrum, it appears that the scalp is the most common pulling site in adults, adolescents, older children, and younger children (Franklin et al., 2008, 2011; Walther et al., 2014; Woods et al., 2006). Pulling can be both automatic (i.e., outside awareness) and focused (i.e., in response to identifiable affective triggers) within each individual, rather than exclusively one form or the other (Flessner et al., 2008a, 2008b), although it appears that there may well be a greater preponderance of automatic pulling in younger samples (Franklin et al., 2010; Panza et al., 2013). The concept of a premonitory urge, which has been discussed extensively in the context of tic disorders (Leckman et al., 1989), also appears to be important in TTM, as most participants in TTM studies to date have reported at least some tension or some other unpleasant sensation that precedes if not precipitates pulling

(Woods et al., 2006). It is possible that young children have not developed the expressiveness skills and emotional awareness to be able to identify or to report such phenomena, which may necessitate an emphasis in behavioral treatment on identification of high-risk times for pulling rather than relying on the patient's ability to recognize and report that the urge to pull is mounting.

What Are the Current Treatment Options?

A wide variety of treatments are attempted clinically to alleviate TTM symptoms in adults, adolescents, and children, including cognitive and behavioral therapies, supportive counseling, support groups, hypnosis, medications, and combined approaches. The scientific literature supporting the efficacy of any of these approaches in adults, however, is not well developed, with fewer than 20 randomized controlled trials available to guide treatment choice and implementation. Most of these trials have examined behavioral therapies or medications, and their collective findings have been somewhat mixed, especially with respect to the efficacy of medication. Further, only two of these randomized trials were conducted with pediatric samples (Bloch et al., 2013; Franklin et al., 2011), despite clear evidence that TTM is a pediatric onset disorder. Where this leaves us as a field is with information about treatment outcome derived almost exclusively from adult samples, which may underestimate the potential role of developmental factors on treatment process and outcomes. Nevertheless, we view it as wise to make use of the observations from adult studies to help guide treatment of youth, provided of course that clinicians are fully aware of the caveats they need to keep in mind when describing the empirical support of their recommendations to patients and families.

Although a comprehensive review of the entire treatment literature in adults is beyond the scope of the current report, recent reviews (e.g., Chamberlain et al., 2009), as well as our own review, highlight several key points: (1) cognitive-behavioral treatments are associated with relatively large effect sizes in adults following acute treatment, although relapse appears to be a problem; (2) selective serotonin reuptake inhibitors (SSRIs) generally do not appear to be efficacious in reducing hair pulling symptoms per se; (3) several compounds that appear to affect other neurotransmitter systems hold some promise for the treatment of TTM; (4) combined treatments with behavioral therapy plus medication may also prove useful; and (5) the absence of evidence from randomized controlled trials conducted with pediatric samples hinders treatment development and treatment planning for perhaps the most vulnerable population of TTM sufferers. In our review of the treatment literature, we endeavor to provide information from adult treatment trials as well, given that the randomized evidence in children and adolescents is so scarce.

The behavioral interventions for TTM used across the developmental spectrum have generally included three core elements, each of which were included in Azrin and Nunn's (1973) initial clinical trial on behavioral treatment for TTM and other "nervous habits": (1) awareness training, wherein techniques (e.g., self-monitoring) are implemented to improve the patient's awareness of pulling and, better yet, the patient's awareness of the urge that precedes pulling; (2) stimulus control, which includes a variety of methods that serve as "speed bumps" to reduce the likelihood that pulling behavior begins; and (3) competing response training, where patients are taught at the earliest sign of pulling or of the urge to pull, to engage in a behavior that is physically incompatible with pulling for a brief period of time until the urge subsides. These core methods comprise the main elements of contemporary behavioral treatment, although some habit reversal training protocols (e.g., Rothbaum & Ninan, 1994) have also included other techniques (e.g., relaxation training, cognitive strategies to address dysfunctional thoughts that precipitate pulling).

Expert opinion (e.g., Flessner et al., 2010) is convergent with the treatment outcome literature in supporting the use of cognitive-behavioral treatments that include habit reversal training as the first-line option in TTM. It is also generally

accepted now that selective serotonin reuptake inhibitors (SSRIs), though potentially useful to address comorbid symptoms of anxiety and depression, are not considered first-line treatments for pulling per se. One study supported the efficacy of an SSRI in combination with behavioral therapy over behavioral therapy and medication alone in adults (Dougherty et al., 2006); replication of these findings is needed. New developments in pharmacotherapy discussed below open the possibility for examining the relative and combined efficacy of novel approaches in concert with behavior therapy as well. Whether these treatments should be started simultaneously or delivered sequentially – for example, premedication with an agent of established efficacy followed by behavioral intervention when pulling urges are lowered by medication effects – still needs to be evaluated using randomized designs.

Behavior therapy, though efficacious, is not without its limitations, the most pressing of which is the observation that relapse following treatment is common in adult patients (e.g., Lerner et al., 1998). Treatment development work conducted in several labs has examined whether behavior therapy involving habit reversal training can be augmented by methods designed specifically to address negative emotions (e.g., Keuthen et al., 2010, 2012; Woods et al., 2006). Findings from Woods and colleagues' large, randomized controlled trial examining the relative efficacy of acceptance-enhanced behavior therapy (AE-BT) versus a psychoeducation/supportive counseling control condition are about to be submitted; Keuthen and colleagues found that behavior therapy enhanced with emotion regulation methods adapted from the dialectical behavior therapy (DBT) was superior to a minimal attention control group in terms of reducing TTM symptoms and enhancing emotion regulation skills. There is also hope that the research tools developed to examine pulling styles more specifically will aid clinical researchers in providing more targeted behavioral interventions that can be tailored to individual pulling profiles.

Recent developments in pharmacotherapy offer encouragement that therapies which modulate neurotransmitter systems other than serotonin will prove helpful in reducing pulling behavior and pulling urges. Bloch and colleagues' thorough review of the treatment outcome literature (Bloch et al., 2007) highlights the fact that selective serotonin reuptake inhibitors offer very little in the way of clinical benefit above and beyond what can be expected from pill placebo. Clomipramine, a tricyclic antidepressant with serotonergic and other properties, appears to be more efficacious than placebo, but its unfavorable side effect profile renders it a second-line treatment. Instead, new data have emerged to support at least preliminarily the efficacy of an opioid antagonist (naltrexone), a glutamate modulator (N-acetylcysteine), and an atypical neuroleptic (olanzapine) for TTM. A summary of each is provided below.

Two published studies have examined the effects of naltrexone on pulling behavior; the logic of its use is that TTM appears to be appetitive, and some investigators have emphasized the phenomenological and underlying neurobiological overlap with other forms of addictive behavior (e.g., Grant et al., 2007). Accordingly, medications that block opioid binding may well prove useful in decreasing the positive reinforcement derived from pulling, hence decreasing urge strength and affecting the behavior. An open-label study on fourteen children with TTM found that naltrexone reduced hair pulling urges and behavior and was not associated with any significant side effects (de Sousa, 2008). However, Grant and colleagues (2012) failed to find a difference between naltrexone and pill placebo in a randomized controlled trial in adults with primary TTM. To date, then, there has yet to be a positive peer-reviewed, double-blind study of naltrexone in individuals with TTM, which compromises assessment of its potential usefulness in clinical practice. Further study of the efficacy and safety of this intervention is needed, as is more basic research on its mechanism of action.

Formal if not functional similarity between the repetitive behaviors seen in tic disorders and those seen in TTM led other neurobiologically oriented investigators to examine the potential utility of atypical neuroleptics to treat hair pulling, either alone or in combination with SSRIs. In

the first randomized, controlled study of this intervention, monotherapy with the atypical neuroleptic olanzapine was found superior to pill placebo in adults (van Ameringen et al., 2010), although the potentially significant side effect profile for this class of medications continues to render them a second-line option when other treatments are available or have not been attempted in a given patient.

Perhaps the most important recent development in pharmacotherapy for TTM involves the use of the glutamate modulator NAC, which was found superior to pill placebo in a randomized controlled trial for adults with TTM (Grant, Odlaug & Kim, 2009). Treatment response rates for the NAC condition were not only clearly superior to the control condition, but they also yielded rates that were comparable to those observed in CBT trials with adults. Further, the side effect profile was quite favorable, which may well make this compound the most promising recent development in the field. Notably, NAC is not an FDA-regulated product, so it is readily available in health food stores. Comparability of products containing NAC from manufacturer to manufacturer, however, is unknown.

Building upon this encouraging foundation, Bloch et al. (2013) conducted a randomized, double-blind, placebo-controlled trial examining the efficacy of NAC for pediatric TTM. Unfortunately, NAC failed to separate from PBO at posttreatment on any primary or secondary outcomes, and the modest rate of clinical responders (25% for NAC and 21% for PBO) indicated that the failure to separate was not due to a floor effect in the PBO condition. Clinically we encounter many child and adolescent patients who come to our open clinic already taking NAC; we have not had the opportunity to systematically study their phenomenology and treatment response.

Finally, in the only published randomized controlled trial ever published examining the efficacy of behavior therapy for pediatric TTM, Franklin and colleagues (2011) found that the behavioral intervention was superior to a minimal attention control condition at posttreatment; moreover, in contrast to the long-term outcomes in adults, pediatric TTM patients who received BT maintained their gains through a 40-week follow-up period. In a sub-analysis of data from this trial, it appeared that younger children (ages 7–9) who completed behavior therapy were more robust responders than their older counterparts. The very small sample size for the behavioral condition ($N = 12$) precludes strong conclusions, but this outcome is convergent with the clinical supposition that younger patients, whose TTM may be more automatic, less affectively driven, and less comorbid with other psychiatric illnesses, are more likely than older patients to respond to the core interventions in habit reversal training, which include self-monitoring, stimulus control, and competing response training.

Recommendations Regarding Existing and New Clinical Strategies

Significant advances have been made over the last decade in TTM research, and we now have additional information on TTM's prevalence (e.g., Hajcak et al., 2006), the functional impact and effectiveness of treatments available in community settings (Franklin et al., 2008; Woods et al., 2006), TTM's core psychopathology (Flessner et al., 2007, 2008; Panza et al., 2013), the collective opinions of treatment experts regarding clinical management of TTM (Flessner et al., 2010), potential utility of various combined treatment approaches (Dougherty et al., 2006; Keuthen & Piacentini, 2012), and the development and empirical evaluation of novel pharmacological approaches that hold promise for clinical care while simultaneously informing us about TTM's underlying neurobiology (e.g., Chamberlain et al., 2007, 2009; Bloch et al., 2007). This wealth of new information has advanced the field considerably with respect to TTM assessment, improved our understanding of TTM's phenomenology, and put us collectively in a better position to evaluate the treatments that are available thus far. At the same time, there is still much to be learned, especially in the areas of pediatric TTM presentation, longitudinal course,

impact upon families, and the creation of a developmentally sensitive treatment that flexibly takes into account the child's awareness of pulling, capacity for understanding the precursors to pulling, ability to provide sustained attention, and willingness to postpone reinforcement (positive or negative) toward the broader aim of reducing pulling urges down the line by refraining from pulling in response to urges now.

The data on pulling styles may be especially important clinically, appears to vary across the developmental spectrum, and likely reflects different affective functions of pulling that need to be taken into account when devising treatment strategies. Automatic pulling, or pulling that takes place outside of awareness and often in the context of sedentary activities, appears to be highly responsive to tactile antecedents (e.g., touching head with fingertips), whereas focused pulling seems to be more responsive to affective or cognitive antecedents. The focused vs. automatic pulling differentiation should not be viewed as dichotomous pulling subtypes, however, as it also appears that most individuals engage in both forms of pulling. However, the preponderance of automatic or focused pulling is important to identify as this has treatment implications, as does the context in which form is more likely to occur. Many experts believe that automatic pulling may be more responsive to the behavioral techniques that comprise habit reversal training, most notably awareness training (increasing the patient's awareness of the environmental and tactile antecedents of pulling episodes), stimulus control (making the environment less conducive to pulling), and competing response (engaging in a behavior that is physically incompatible with pulling in response to urges to pull). Focused pulling, on the other hand, may also require techniques that address affective and cognitive antecedents more directly, such as those offered in dialectical behavior therapy (Keuthen et al., 2012) and acceptance and commitment therapy (Woods et al., 2006). An open clinical trial of DBT provided preliminary support for the efficacy of DBT-enhanced CBT in treating adults with TTM (Keuthen et al., 2010); follow-up of patients included in that study indicated that gains were generally maintained in the active treatment group at both 3- and 6-month follow-up (Keuthen et al., 2011). The randomized trial that followed provided further evidence for the efficacy of this approach (Keuthen et al., 2012) and a solid foundation from which to move forward in terms of the utility of this combined approach.

TTM in children and adolescents may well be governed by a greater preponderance of automatic pulling (Flessner et al., 2007, 2008; Franklin et al., 2008; Tolin et al., 2007), which could help explain why initial reports of the efficacy and durability of habit reversal training are more promising in younger samples (Franklin et al., 2011; Tolin et al., 2007). With a larger sample size, scientists will be able to explore this more formally by directly comparing the relative outcomes of children and adolescents with a preponderance of automatic pulling to those with a pulling profile characterized by more focused pulling. Such a study has now recently been completed and is described in detail elsewhere (Weiss & DiLullo, 2009): sixty children and adolescents were randomly assigned to treatment with behavior therapy involving habit reversal training or to a comparison condition consisting of psychoeducation and supportive counseling, which replicates the design used by Keuthen et al. (2012) to examine the DBT-enhanced form of HRT. Data from the MIST-C was collected to document the pulling styles of each participant in the trial, which will then allow a direct comparison of the preponderance of focused or automatic pulling in order to determine whether pulling style predicts outcome (regardless of treatment assignment) or moderates outcome (affects one treatment condition more than the other one).

Prognosis

As indicated in the discussion of current treatment options, people with TTM should receive CBT, more specifically habit reversal training, which includes the components of awareness training, stimulus control, and competing

response training. As lamented above, the lack of evidence from randomized controlled trials in younger populations until very recently (Franklin et al., 2011) makes it difficult to draw confident conclusions about outcome and thus use data to drive clinical decision-making. Our ongoing treatment study focusing on HRT versus a psychoeducation/supportive counseling control condition in youth with TTM will improve our understanding of CBT for pediatric TTM and will provide much needed information to improve the efficacy and durability of behavioral interventions. With respect to evaluating the usefulness of pharmacotherapy in treating TTM, randomized controlled trials of NAC and other promising therapies both alone and in combination with behavioral treatments are still needed across the developmental spectrum, although the Bloch et al. (2013) study described above dampens enthusiasm for examining NAC per se. It may be the case, however, that combined treatment will allow for increased symptom improvement and less severe relapse rates than behavioral treatment alone; we cannot be certain of this however until the field conducts a greater number of efficacy trials on this topic.

Our expert opinion regarding treatment options must include the caveat that although CBT is the first-rate treatment for TTM, this line of treatment is as yet not readily accessible to TTM sufferers across the developmental spectrum. Many clinicians who treat individuals with TTM do not practice CBT nor do they incorporate HRT into their treatment. Dissemination efforts must increase in order to allow this treatment to become more commonly practiced in community settings. Currently, the gains made in TTM research concerning assessment and treatment effectiveness have not impacted clinical practice beyond the academic context. A recently completed study of stepped care in the treatment of adult TTM (Rogers et al., 2014) may offer another road forward: in this trial, Step 1 comprised 10 weeks of web-based self-help via StopPulling.com, which was compared to a waitlist control. Participants were then offered Step 2, which was an 8-week course of HRT. Step 1 was superior to waitlist, and over a third of those who entered Step 2 made clinically significant improvements in self-reported hair pulling. This approach may allow for a more efficient use of clinical resources and may well help reduce waitlists for clinical services in areas where HRT expertise is available.

In light of the information chronicled above, it appears that the typical patient presenting with TTM has several empirically supported treatments to consider, although the strength of the evidence base precludes drawing especially confident conclusions about outcome. The evidence is especially sparse in support of treatments for pediatric TTM in particular, although there are some encouraging signs that this problem can be addressed using the core techniques of HRT. TTM appears to become more complex and comorbid over time, which in our view presents a strong argument for trying a course of behavioral treatment before such complexities become more readily apparent. This is not to say that toddlers with TTM would necessarily be good candidates for this form of treatment, as skills such as self-awareness and ability to monitor one's own behavior may be developmentally beyond what most children in this age range can manage. We await more research on the effects of clinical strategies that are brought to bear to help young children refrain from pulling (e.g., placing stuffed animals with them when they attempt to fall asleep, keeping hair pulled back or cut short).

Summary and Recommendations

Much has been accomplished in the last 10 years with respect to TTM research, and we hope that this work has set the stage for the next generation of TTM researchers to further advance our knowledge regarding TTM's core psychopathology, TTM's underlying neurobiology, TTM's responsiveness to existing treatment, empirically informed treatment development, and dissemination of the most effective methods into community settings where patients and their families can access them. Instrument development efforts have helped lay the foundation for such research, as we now have have

psychometrically acceptable measures of TTM severity and pulling styles across the developmental spectrum, which will enable researchers to document symptom severity and symptom change and to examine what may well be a critical predictor if not moderator of treatment outcome, namely, the preponderance of automatic versus focused pulling. There have been improvements in clinical trial designs in this period as well, such that there are now more randomized studies available to help estimate the likely effects of given treatments in clinical settings. Certainly, though, we are not close to where we need to be as a field to arrive at a point where clinicians will be able to convey with great confidence that TTM treatment is either universally or completely effective for all. More randomized controlled trials will be needed to replicate the recent findings with behavior therapy for children and adolescents, for combined treatment in adults, and for NAC, atypical neuroleptics, and opioid antagonists across the developmental spectrum. Given that the evidence from treatment studies indicates that treatment response to any of the available therapies is neither universal nor complete, it is also clear that the development of new pharmacotherapies opens up the possibility of studying how these approaches can best be combined with behavioral interventions. The clinical management of partial and nonresponse also needs to be addressed – in this case the OCD literature provides useful guidelines for ways to combine efficacious treatments in order to move patients closer to subclinical status (e.g., Franklin et al., 2011; Simpson et al., 2008). It is also imperative to focus efforts on improving the durability of treatment gains given that relapse appears to be common even in those adults who have received adequate treatment and responded well to it initially.

The promise of NAC in adults may well prove to be strongest signal yet in the pharmacotherapy literature, yet its lack of efficacy in youth gave researchers pause in endorsing its broader use (Bloch et al., 2013). Questions about its mechanism of action must also be raised in the context of subsequent NAC treatment trials; such studies may also enhance our understanding of TTM's complex neurobiological underpinnings. Studies of long-term efficacy and safety are also needed. Because NAC appears to be both efficacious and tolerable in adults, it continues to stand out among the various medication candidates for direct comparison with CBT and to combined treatment in future trials. Bloch and colleagues provide several reasons why they believe that NAC was not efficacious in youth – these hypotheses must also be tested empirically before giving up on this potentially useful, safe, and readily accessible form of intervention for youth with TTM.

Despite a decade's worth of substantive progress in TTM research, the reality remains that the impact of this work on clinical practice remains minimal outside the academic context, and this stands as the next if not the largest challenge still facing the field. TTM is likely not alone with respect to this state of affairs: investigators have noted that empirically supported cognitive-behavioral treatments for a wide variety of disorders are often not available in community settings and, when accessed in such settings, are often delivered sub-optimally (Shafran et al., 2009). We recognize this as a more general problem that faces the field broadly, but our experience clinically and in conducting treatment trials for TTM suggests that it may be especially acute in TTM. Families have contacted our clinic to participate in our TTM research trials from outside of our region and even nation and report doing so because they have exhausted local efforts to find a treatment provider that has even minimal expertise with TTM. Efforts to improve awareness of TTM must be accelerated in order to assist providers in developing more basic awareness, knowledge, and competence in TTM and its treatment. We are pleased to see that patient-oriented organizations devoted to the dissemination of information about anxiety disorders (e.g., Anxiety Disorders Association of America) and obsessive-compulsive disorder (e.g., Obsessive Compulsive Foundation) have been working with the leading patient-oriented organization in TTM (Trichotillomania Learning Center) in attempts to include more presentations

about TTM to their respective memberships in the last 5 years. Such efforts are critical in getting mental health practitioners more directly involved in the process of providing treatments for TTM. Identifying sources for research funding to support these efforts is also of paramount importance, as it is difficult to extend the reach without such support. There are at present many encouraging avenues to pursue using implementation science methodology to train mental health providers and help foster cultures in community mental health settings that will permit uptake of the clinical approaches that have proven most promising in the efficacy context (see Beidas et al. & Glisson et al. for examples). It remains our hope that a lack of awareness of this low base-rate condition does not prove to be an unbreachable barrier in making effective treatments more widely available.

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Part VII

Internalizing Disorders: Trauma and Stress Related Disorders

Attachment Disorders

13

Kathleen T. Baynes and Thomas G. O'Connor

The American Psychiatric Association's DSM-V made several notable changes to the diagnostic entity of "attachment disorders.[1]" These changes were anticipated from a growing evidence base and create a stronger agreement with the ICD-10 – and imply a firmer basis for characterizing the disorder(s). This chapter focuses on diagnostic progress and those matters that remain unresolved concerning conceptual models and biological bases, as well as assessment and treatment. Specifically, we first consider attachment disorders from an attachment theory perspective. Second, we report on the diagnostic features and differential diagnoses of attachment disorders. Third, we review briefly some of the main research findings from the collection of studies in this area. Fourth, we review the evidence supporting the use of particular assessment models and practices. Fifth, we review the evidence base on treatment. Sixth, we provide a case example to illustrate some of the clinical decision-making in treating children with an attachment disorder.

[1]In order to accommodate the differences in diagnostic labels within and between diagnostic systems, throughout the chapter we use the term "attachment disorder(s)" to refer to the two forms of reactive attachment disorder listed in DSM-IV and reactive attachment disorder and disinhibited social engagement disorder in DSM-V, as well as parallel diagnoses in the ICD manual.

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Attachment Disorders: Conceptual and Historical Background

A persisting and fundamental question concerning the nature of attachment disorders is the nature of their link with attachment theory that was developed by Bowlby (1982, 1988), Ainsworth (Ainsworth, 1989; Ainsworth, Blehar, Waters, & Wall, 1978), and their colleagues which spawned one of the more influential strains of developmental-clinical research in the past several decades. Probably the strongest argument in favor of conceptualizing attachment disorders alongside attachment theory is the overwhelming evidence (reviewed below) that children with an attachment disorder have been exposed to depriving and neglectful experiences not just in general terms of nutrition and social contact but most particularly in the early child-parent attachment relationship (O'Connor & Zeanah, 2003). In this case, the attachment disorder is a consequence of the prior attachment experiences and, more specifically, the absence of opportunities to form a selective attachment relationship with a caring adult.

On the other hand, there are obvious and important tensions in conceptualizing attachment disorder within even a broad understanding of attachment theory and research. One of the most basic concerns the description of the behavioral phenotypes. Several decades of research on

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attachment theory yield a robust and detailed description of attachment behavior in infants, children, adolescents, and adults according to assessment strategies that use observational, narrative, or interview techniques. Phenotypes from these alternative assessment measures are distinguished as secure or insecure, a designation which describes the degree to which the individual uses the attachment figure as a secure base for exploration (a fundamental distinction derived from observational assessments in children) or articulates a state of mind with respect to attachment relationships that is coherent and emotionally open to attachment-relevant thoughts, feelings, and memories (a fundamental distinction derived from interview assessments with older adolescents and adults). Importantly, the behavioral phenotypes distinguishing secure and insecure in the attachment research literature do not appear in diagnostic criteria: the notions of security and insecurity are fundamentally distinct from the phenotypes or symptoms of attachment disorder described in DSM-V (or in DSM-IV, or the ICD).

One attempt to reconcile or at least clarify differences between behaviors or phenotypes described by attachment theory and attachment disorder is to appreciate qualitative differences in the caregiving experiences associated with them. Attachment theory is concerned with normative variation (which does ordinarily include clinical disturbance in caregiving quality, such as maltreatment), judged in terms of the caregiver's sensitivity and responsiveness to the psychological needs of the child. Caregiving experiences that are consistently and significantly sensitive and responsive lead the child to develop a secure attachment relationship with that caregiver. Conversely, caregiving experiences that are consistently and significantly insensitive and unresponsive lead to the child developing an insecure attachment relationship with that caregiver (Ainsworth, Bell, & Stayton, 1971; Ainsworth et al., 1978). Studies of high-risk samples such as children of parents with significant mental illness or children who experienced maltreatment helped to refine the different subtypes of insecure attachment and particularly to highlight that form of insecure attachment, disorganized, that is most closely associated with maladjustment in the child (Barnett, Ganiban, & Cicchetti, 1999; Cyr, Euser, Bakermans-Kranenburg, & Van Ijzendoorn, 2010; van Ijzendoorn, Schuengel, & Bakermans-Kranenburg, 1999).

In contrast, attachment disorder is associated not with variation in caregiving quality but rather with the absence of an opportunity to form a selective attachment relationship. Significantly, Bowlby (1982) anticipated this qualitative distinction and an incompatibility between attachment research and attachment disorder when noting that attachment relationships form in what he referred to as the "environment of evolutionary adaptedness." Children without opportunities to form selective attachment relationships such as those in institutional settings are arguably not developing within an environment of evolutionary adaptation. Accordingly, the attachment theory that Bowlby developed and that was substantiated by decades of subsequent research may not be applicable to children who did not have opportunities to form normative (secure and insecure) attachment relationships. Research supporting this fundamental distinction is outlined in a subsequent section. It is worth anticipating here that the methods developed from and for research on secure and insecure attachment relationships may not be suitable for children with an attachment disorder. That is, as discussed below, results from several studies show that, when children with a bona fide or strongly suspected attachment disorder are assessed using attachment research methods, contrary and puzzling findings may result. Discussions of attachment theory and attachment disorder are notorious for misstatements and misunderstandings. These may be allayed to the extent that there is a clear description of the behaviors of interest, an account is given of the context in which the behaviors were observed or reported, and some consideration is given to the quality of caregiving experiences which focuses particularly on the extent to which the child had opportunities to form a selective attachment relationship.

An alternative view is to regard the term "attachment disorder" as something of a misnomer. That is, "attachment disorder" may be better understood as a neurodevelopmental disorder in terms of fundamental disturbances in the expression and understanding of emotion, disturbances in pragmatic language, and problems in cognitive inhibition and attention and executive function, among other disturbances. According to this alternative model, the social and relationship disturbances observed in children with an attachment disorder are secondary to fundamental problems in socio-emotional and cognitive development which resulted from the early deprivation and neglect, rather than an expression of features associated with attachment disorder. In this more neurodevelopmental model, disturbances in attachment relationships are considered as one among many of the social, cognitive, and relationship disturbances associated with severe deprivation and neglect. The emphasis here is on delineating the "component parts" of the developmental disturbance resulting from neglect as opposed to attributing deficits to an omnibus concept, such as attachment, which may not adequately reflect or capture all of the clinical phenomena – or be the essential cause of all of the evident disturbances. This distinction is not merely academic, as it implies potentially very different manners of assessment and treatment. For example, if impairments in peer relations are evident, then it may be helpful to apply social skills training (SST) as a component treatment rather than presume that progress in peer relationships will parallel clinical progress in the quality of the child's attachment relationships with caregivers.

Attachment Disorders: Diagnostic Features and Differential Diagnosis

Core features of attachment disorder have been reliably described for the past 75 years; most of the clinical attention was on what is now referred to as disinhibited social engagement disorder (see Table 13.1). Specifically, children who experienced caregiver deprivation have been described as "superficially affectionate" and "indiscriminately friendly" as well as superficially affectionate and showing "affectionless psychopathy" (Goldfarb, 1943; Levy, 1937; Provence & Lipton, 1962; Tizard & Hodges, 1978; Tizard & Rees, 1975; Wolkind, 1974). The consistency with which very many authors described this phenotype and the obvious impairment associated with it led to the introduction of reactive attachment disorder in the DSM-III (APA, 1980). This early definition focused on symptoms observed in infants and included failure to thrive and a lack of social responsiveness and relatedness. A diagnosis also required that there was a gross lack of care, specifically emotional neglect or social isolation. Subsequent diagnostic modifications (DSM-III-R, 1987) eliminated growth failure and broadened the onset of observed behaviors to within the first 5 years, after the age at which clear attachment behavior is expected and when functional impairment can be observed across a range of social contexts. The disturbed social relatedness that was the hallmark of the disorder was noted to reflect either a failure to engage in social interactions or an indiscriminate sociability. These two subtypes were more clearly demarcated in DSM-IV and DSM-IV-TR (APA, 1994, 2000). Children with the "inhibited" subtype were described as withdrawn or ambivalent across most social interactions; they also failed to seek out the attachment figure when it would be developmentally expected. Children with the "disinhibited" subtype displayed lack of selectivity and excessive familiarity toward others (including strangers) and did not reliably seek out the caregiver for assistance or comfort when it would be developmentally expected.

Several kinds of criticism were raised about the diagnostic criteria. One basic problem was that, until arguably the DSM-IV, there were not published studies using the diagnostic criteria (or validated assessment approaches) for the disorder. Additionally, the diagnostic language was problematic in many other ways that implied a misunderstanding of attachment. For example, as part of the DSM-IV criteria for the disinhibited form of reactive attachment disorder, there was reference to "diffuse attachments" – and oxymoron given that attachments

Table 13.1 Diagnostic features of attachment disorders according to alternative manuals

	DSM-III	DSM-III-R	DSM-IV and DSM-IV-TR	DSM-V-RAD	DSM-V-DSED	ICD 10
Key diagnostic criterion/ criteria	Lack of social responsivity	Disturbed social relatedness	Disturbed social relatedness	Inhibited emotionally withdrawn behavior	Overly familiar behavior with strangers	Persistent abnormality of social relationships
Onset	Within 8 most	Within 5 years	Within 5 years	After 9 mos, before 5 yrs	After 9 mos, before 5 yrs	Before age 5
Etiology	Lack of care; gross emotional neglect; imposed social isolation	Grossly pathogenic care	Pathogenic care	Extreme insufficient care	Extreme insufficient care	Known history of abuse or neglect
Subtype	None	Withdrawn and indiscriminate	Inhibited and disinhibited	None	None	Reactive attachment and disinhibited attachment
Exclusion	Not due to a physical disorder, mental retardation, or infantile autism	Not due to mental retardation or PDD	Not due to developmental delay or PDD	Criteria not met for autistic spectrum disorder	Criteria not met for autistic spectrum disorder	Criteria not met for PDD

are not, by their nature, "diffuse." Other features attracted criticism, such as requiring the putative cause – a history of severe deprivation or neglect – in the diagnostic criteria. Still other features, such as ruling out developmental delay or pervasive developmental disorder, were not particularly controversial but nevertheless derived from an uncertain evidence base.

Compared with what was available at the time of previous diagnostic manuals, the research evidence base on attachment disorders was relatively substantial for DSM-V. The extent to which this stronger evidence base altered the form of the diagnosis is therefore a matter of some interest. Certainly the most notable change from DSM-IVR to DSM-V is the creation of two subtypes of disorder, reactive attachment disorder (RAD) and disinhibited social engagement disorder (DSED). RAD constitutes the inhibited form from previous manuals and is characterized as a pattern of "markedly disturbed and developmentally inappropriate" attachment behaviors that are underdeveloped or absent, nonselective, and ultimately unsatisfying. Disinhibited social engagement disorder, which is the inhibited form from previous manuals, is characterized by a pattern of overly familiar and superficial social behavior. Each of these disorders continues to have a prerequisite of extreme insufficient care, and the need to consider developmental delay and autism spectrum disorder also remains.

Diagnosis of attachment disorder in older children is challenging because the core symptoms are based on those features observed in young children. The degree to which the core symptoms of RAD and DSED, which were initially described in young children, apply equally well to older children and adolescents is uncertain. Nonetheless, as described below, the disorder is thought to be persistent beyond early childhood in at least a sizable minority of cases. And, children with an attachment disorder show a range of impairments in peer relationships and many aspects of social, affective, and cognitive development. Quite how, and if, these findings can be integrated into diagnostic definitions for older children remains a central task for further longitudinal clinical research. An additional developmental consideration is that DSM-V advises caution in diagnosing children over 5 years of age who had not previously shown the disturbance. That comports with the virtual absence of clinical and research evidence of an onset of attachment disorder past infancy or early childhood.

Given the changes in diagnostic criteria for RAD/DSED between and within diagnostic system, it is perhaps not surprising that we currently lack reliable population-level data on prevalence. It has been reported in the DSM that the condition is extremely rare in the population. One essential qualifier, however, is that the rate of attachment disorder or behaviors strongly suggestive of disorder vary widely depending on the population studied. So, for example, virtually all studies of children who experienced institutional care commonly report symptoms of attachment disorder behavior (although virtually none has used formal diagnostic criteria); findings from this set of studies is elaborated below. Children in foster care are also thought to be at high risk for attachment disorder symptoms; one study reported a prevalence in a foster care sample of 19% (Lehmann, Havik, Havik, & Heiervang, 2013). It is expected that the rate of attachment disorder would be exceedingly rare in nonselected or general population samples. Few studies of that kind are available, but it is notable that one study suggested a rate of 1% in a socially deprived sample (Minnis et al., 2013a). Methodological concerns regarding diagnosis, discussed below, mean that these rates must be considered with caution, but there is growing recognition that the condition may not, in some clinical contexts, be very rare.

The differential diagnosis for attachment disorder requires a broad array of considerations and complicated – which is to say somewhat speculative – propositions (Table 13.2). Many different problems can arise from early adversity and may therefore accompany or confound a diagnosis of RAD or DSED. Rates of co-occurring disturbances vary to widely too provide reliable estimates, and the pattern of co-occurring symptoms may vary across populations, e.g., children from grossly abusive and

Table 13.2 Differential diagnosis and distinguishing features

Alternative Diagnosis/ classification	Overlapping symptom or behaviors	Potential distinguishing features and assessment strategy
Intellectual disability	Cognitive and language delay, problems in executive function, cognitive inhibition	Abatement of symptoms may occur in children with RAD/DSED, perhaps associated with stability in caregiving environment Diagnosis and targeted management are warranted if symptoms are met
Autistic spectrum disorder/ pervasive developmental disorder	Stereotyped behavior; social relationship impairments; lack of social reciprocity; deficits in mentalization	Abatement of symptoms may occur in children with RAD/DSED, perhaps associated with stability in the caregiving environment Children with RAD/DSED may lack family history of autism/PDD
Attention deficit hyperactivity disorder	Attention, impulsivity	ADHD symptoms in RAD/DSED may be less pervasive across setting and may be exacerbated in attachment-related contexts ADHD diagnosis and targeted treatment are warranted if symptoms are met
Post-traumatic stress disorder	Increased hypervigilance, nightmares, anxiety	Symptoms in PTSD may be more episodic, and the child may gain some relief from symptoms with a preferred caregiver PTSD diagnosis and targeted treatment are warranted if symptoms are met
Disruptive behavioral disorder (ODD or CD)	Disruptive behavior, defiance, lying, callous/unemotional behavior, lack of prosocial emotions	Hostile attribution bias and other cognitive distortions associated with disruptive behavior may be lacking in children with RAD/DSED ODD or CD diagnosis and targeted treatment are warranted if symptoms are met
Insecure attachment	Relationship conflict; attachment figure is unable to assuage child's distress	Children with an insecure attachment will display evidence of seeking out caregiving when distressed, even if done in an avoidant, ambivalent, or disorganized manner; history of severe caregiving deprivation/neglect is required for RAD/DSED diagnosis Attachment research measures may not be able to distinguish insecure attachment from RAD/DSED

neglectful backgrounds compared with children who experienced institutional privation. Commonly reported co-occurring disorders and conditions include intellectual delay, ADHD, oppositional and conduct disorder, anxiety and depression, and trauma; somewhat less common but nonetheless striking are symptoms of autistic-like behaviors. Some of the most widely discussed co-occurring symptoms and disorders that have proven challenging for making diagnosis of RAD/DSED are provided in Table 13.2, along with attachment insecurity.

To a considerable extent, problems in comorbidity merely indicate that multiple kinds of disorders, including attachment disorder, may result from a history of severe caregiving deprivation or neglect. For example, intellectual disability and developmental delays can result from a poor early caregiving environment and have been widely reported in children with attachment disorder symptoms. There is no doubt that making the additional diagnosis of intellectual disability will be required in many cases, and whether or not these symptoms can been seen as primary or secondary to attachment disorder will be difficult to disentangle. What is notable is that cognitive gains made following change in caregiving environment (particularly moving from an institu-

tional to family setting) have been reported (Beckett et al., 2006; Fox, Almas, Degnan, Nelson, & Zeanah, 2011), and in these cases it is of considerable interest if these gains are coordinated with improvements in attachment and social relations.

An attachment disorder is specifically ruled out if the child meets criteria for pervasive developmental disorder or autistic spectrum disorder. Although abnormalities in social and emotional reciprocity and social communication difficulties are found in both disorders, it is possible to distinguish these conditions because children with RAD/DSED do not show the full array of autistic symptoms. Several reports suggest that a minority of children who experienced institutional deprivation exhibit "quasi-autistic" symptoms (Levin, Fox, Zeanah, & Nelson, 2015; Rutter et al., 1999). In this case, clinical data on developmental course may be helpful as a useful differentiating factor, as some children whose autistic-like symptoms may be caused by institutional rearing show significant clinical improvement that would not be expected in typical autism (Rutter et al., 1999).

Early adversity and experiences with maltreatment also put children at risk for post-traumatic stress disorder, anxiety disorders, disruptive behavior disorders, and attention deficit hyperactivity disorders. For example, there is now growing evidence that ADHD and some of its neurocognitive correlates are associated with deprivation (Kreppner, O'Connor, & Rutter, 2001; Slopen, McLaughlin, Fox, Zeanah, & Nelson, 2012). Each of these disorders may be comorbid with attachment disorders, but their diagnostic criteria are distinct and, if met, require a separate diagnosis. Subjugating standard diagnostic symptoms of inattention, disruptive behavior, anxiety, or other mental health disorders to an attachment disorder diagnosis has been proposed by some writers, but there is no clinical research evidence to support this practice. Furthermore, as reported below in the case example, symptoms of these disorders may benefit from treatment (e.g., medication for ADHD) independently from progress or lack of progress in managing symptoms of attachment disorder.

Empirical Research Findings

Research findings reported in the past decade substantiate and extend considerably early clinical reports in several ways. For example, it now seems quite evident that it is the absence of a consistent caregiver in the early years or the lack of opportunity to form a selective or discriminating attachment relationship that has a causal role in the development of attachment disorder. An important supporting finding is that reports of bona fide attachment disorder symptoms are extremely rare in children from "typical" high-risk settings – that is, children who experience poor caregiving such as maltreatment but nonetheless have opportunities to development selective (albeit insecure) attachment relationships with a caregiver. What was perhaps not anticipated from earlier writings is the wide variation in attachment outcomes among children who did not have opportunities to develop selective attachment relationships. For example, in the English and Romanian Adoptee (ERA) study, approximately 30% of children who experienced 24–42 months of institutional care exhibited a severe disinhibited disturbance, which persisted longer term in a further sizable subset (O'Connor & Rutter, 2000; Rutter et al., 2007). The finding that it is only a minority of children who display evidence of an attachment disorder following prolonged institutional care has been replicated in other studies. On the other hand, these same studies also indicate that the disturbance is persistent. Data from the ERA study which assessed children at 4, 6, and 11 years of age is typical of the observational follow-up studies in documenting considerable stability in individual differences and a persistence of disinhibited behavior long after removal from the depriving environment and despite many years of living in a caring and responsive family environment.

Two other findings from several decades ago about etiology are still relevant and significant. One concerns developmental timing or exposure. In his study of "affectionless psychopathy," Wolkind (Wolkind, 1974) found that disinhibition toward strangers was associated with *early* institutional care. The second finding concerns

the nature of the depriving experience predictive of attachment disorder. In contrast to the recent flurry of studies of ex-institutionalized children who experienced global severe deprivation, Tizard and colleagues (Tizard & Rees, 1975) studied children in institutions who had adequate nutrition and cognitive stimulation, i.e., non-global deprivation. They showed that the attachment disturbance was found in children who experienced institutional care even if where adequate care other than attachment was available. That is, the key observation is that attachment disorder symptoms do not constitute an "institutional syndrome" but more specifically a disorder associated with caregiver deprivation. In fact, improving the child-caregiver ratio and opportunities for regular care is associated with a decrease in disinhibited disturbances (Smyke, Zeanah, Fox, Nelson, & Guthrie, 2010; Smyke et al., 2012). Intervention studies, of which the most renown is the Bucharest Early Intervention Project (BEIP; cited above), document intraindividual improvements in attachment relationships and social functioning and also provide strong causal evidence for the role of caregiving manipulations and hint at sensitive periods for these interventions.

The biological correlates of attachment disorder suggest the involvement of several candidates, although progress in understanding the biological bases of attachment disorder behavior from the perspectives of etiology, assessment, and treatment remains limited. Findings from studies using genetics (Drury et al., 2010), neuroendocrine, and neuropeptide targets (Koss, Hostinar, Donzella, & Gunnar, 2014; Wismer Fries, Ziegler, Kurian, Jacoris, & Pollak, 2005) suggest some interesting leads, and this line of study will constitute an important avenue for further investigation. A particularly active area of research into the biological correlates of attachment disorder uses brain imaging techniques. One of the earliest reports was from Chugani and colleagues (Chugani et al., 2001) who described reduced glucose metabolism in children adopted into the USA following institutional deprivation and compared results with adult and child epileptic comparisons. Affected brain regions associated with institutional deprivation included the orbital frontal cortex, prefrontal cortex, and medial temporal structures that include the amygdala and hippocampus. One of the leading neuroscience findings in this area is the observation of greater amygdala volume associated with institutional deprivation (Tottenham et al., 2010). That finding is notable in light of the anxiety symptoms that have been reported. There are, in addition, several other reports focusing neuroscience findings using brain imaging methods. More recent reports using different imaging techniques have expanded the list of brain measures linked with deprivation, including gray matter volume (Sheridan, Fox, Zeanah, McLaughlin, & Nelson, 2012) and reduced cortical thickness (McLaughlin et al., 2014).

Emotional recognition and understanding, core components of effective social interactions with caregivers and peers, have been extensively studied in several groups who have followed children following early institutionalization. Deficits in emotion understanding and emotion labeling have been widely reported (Fries & Pollak, 2004) as have deficits in emotion regulation and symptoms of anxiety (Tottenham et al., 2010); there is also some suggestion that emotion recognition may play a role in the persistence and/or worsening of internalizing symptoms (Colvert, Rutter, Beckett, et al., 2008a).

A related and equally active area of research examines cognitive processing, including executive function, attention, and related neuropsychological constructs. Here, too, deficits have been widely reported, with some evidence of persistence where multiple measures have been taken (Loman et al., 2013; McDermott et al., 2013; Merz, McCall, Wright, & Luna, 2013).

To a considerable extent, the findings so far reported are unable to differentiate the biological effects of severe early deprivation from those that may be particular to the development of an attachment disorder. That is because, in many cases, the predictor has been institutional history rather than that subset of children who experienced institutionalization and exhibited significant attachment disorder symptoms. That is a significant consideration and point of clarification because deficits in cognitive and emotional

development such as executive function may be significantly associated with early deprivation but may not mediate the impact of early deprivation on attachment disorder behavior (Colvert, Rutter, Kreppner, et al., 2008b). Interesting, another study of ex-institutionalized children found that disinhibited social approach that is the characteristic feature of DSED was not related to overall IQ but was related to inhibitory control (Bruce, Tarullo, & Gunnar, 2009).

Research findings on the attachment disorder phenotypes and social, cognitive, and neuroscience correlates beyond early-middle childhood are especially rare. That imposes important limits on the conceptualization and measurement of the disturbance – and of course all matters concerning treatment. One of the few studies of older children was reported by Kay and Green (Kay & Green, 2013) in a sample of looked-after children in the UK; they offer some of the only evidence that the core symptoms of RAD and DSED may have clinical application in older children and adolescents. What is needed now are studies that extend the focus on social and biological correlates into the older age range.

There is some doubt about how well findings related to attachment disorder derived from studies of ex-institutionalized children will extend to populations of children who experienced noninstitutional deprivation, most notably children in foster care, who constitute the largest population of interest for attachment disorder. Studies of children in foster care reliably show that severe attachment problems – including attachment disorder – are comparatively common and may account for some the quite disproportionate level of care and cost in this population (Lehmann et al., 2013; Minnis, Everett, Pelosi, Dunn, & Knapp, 2006). The broader social care context for older children and adolescents was recently discussed in a report which indicates that concerns about depriving caregiving experiences may extend well beyond the case of early institutional privation (Dozier et al., 2014).

A final reference is needed to the small but potentially valuable set of studies that examine attachment disorder symptoms in nonselected (or at most high-risk according to general psychosocial criteria) samples, that is, community samples in which rates of foster care and institutional care experiences (as well as the kinds of depriving care experiences specifically mentioned in diagnostic criteria) would be minimal. What is notable about these studies (Minnis et al., 2013b; Minnis et al., 2007; Pritchett, Pritchett, Marshall, Davidson, & Minnis, 2013) is the finding that the rates of attachment disorder-related symptoms is not as exceedingly rare as the DSM has been suggesting. It may be premature to calculate prevalence rates from these studies, given the limitations of the methods used and amount of detail (e.g., regarding care experiences) that was available, but it is notable that behaviors related to attachment disorder may be endorsed in a broader population of children than is now typically presumed.

Assessment Strategies and Techniques

Assessment of psychopathology in the young child is challenging for multiple reasons. Caregiver reports and patient presentations may be inconsistent, and child development is constantly evolving. As with any other clinical condition, assessment of attachment disorders requires a gathering of history, collateral sources of information about the patient's early caregiving environment, and the patient's engagement with that environment. There are, however, some features of the assessment of attachment disorders that may be particular. One of these is the evaluation of the attachment relationship that the child has with the primary caregiver, by which we mean specific evidence that the child actively seeks comfort from the parent when distressed. Assessing quality of family relationships is, of course, basic in any assessment of childhood psychopathology; what is distinguishing here is the focus on the evidence for attachment behavior specifically. Another particular feature of the attachment disorder assessment is a consideration of the child's engagement with others, that is, the presence of disinhibited social engagement, such as the seeking out of comfort with

strangers or the inappropriate social boundary violations that are the hallmark of DSED.

Quite how these particular assessment challenges may be met has not been completely resolved, although there is progress on several fronts. There are now clinical assessment measures that have been specifically developed to assess attachment disorders in young children. One of the better-known interview measures is the Disturbances of Attachment Interview developed by Zeanah, Smyke and colleagues (Smyke, Dumitrescu, & Zeanah, 2002). Other interview-based measures have been developed and have been found to have at least suggestive construct validity (Minnis et al., 2009; O'Connor & Rutter, 2000). The clear benefit of the interview context is providing the clinician with the opportunity to gather specific detail of behaviors and contexts. Diagnostic criteria have generated considerable debate and not inconsiderable confusion in the professional literature, and that may be reason enough to require detailed behavior data from multiple sources on the child's behavior in multiple settings before rendering a diagnosis.

Questionnaire approaches have also been devised but may not be as widely used or researched. However, questionnaire methods have helped to make assessments of attachment disorder symptoms possible in large-scale surveys (Minnis et al., 2007; Pritchett et al., 2013). Less well developed by perhaps even more valuable are observational assessments, which seek to detect specific behavioral evidence of disinhibited behaviors. Only very limited observational research exists, however. Gleason et al. (Gleason et al., 2011) used a "stranger at the door" paradigm. In an institutional sample, they found a high convergence between a diagnosis of RAD and willingness to leave the home/setting with a stranger. Observational assessments that involve cooperation and experiences of stress, such as asking the caregiver to briefly leave the room, may be valuable opportunities and have been widely suggested (Practice Parameters 2005). The robustness of these observational paradigms is not yet clear, however, and it will clearly be important to assess behavior in more than one context and on more than one occasion.

One of the major lingering assessment questions concerns the use of attachment research measures. It is important to note at the outset that attachment research measures – the strange situation with infants and preschool-aged children, attachment narrative assessments in young school-aged children, and attachment interviews with older children and adolescents – were developed to assess the security/insecurity of an existing attachment relationship. These measures were not designed to assess the attachment disorder question: has the child developed a selective attachment relationship with a particular caregiver? Predictably, then, it might be expected that these attachment research measures will have limitations when applied to the attachment disorder context. Indeed, we now have positive evidence from several studies that attachment research measures show that children with clear symptoms of attachment disorder were nonetheless rated as showing a secure attachment relationship with a caregiver (Chisholm, 1998; Minnis et al., 2009; O'Connor, Marvin, Rutter, Olrick, & Britner, 2003). That implies that attachment research measures may have limited value in determining whether or not a child's behavior meets criteria for an attachment disorder.

Treatment Models and Evidence of Effects

A consistent message from many reviews is that the evidence base on treatments for RAD is severely limited. To the extent that treatment guidelines have been offered by professional bodies (AACAP, APSAC) it is to warn against the use of frankly dangerous intervention approaches. For example, coercive holding treatments have caused injury and death, and there is a general suspicion, based on clinical-anecdotal evidence, that the coercive holding may re-traumatize children. The California Evidence-Based Clearinghouse for child welfare (www.CEBC4cw.org) is a valuable resource for information evaluating the strength of evidence for mental health interventions. The site now

includes information on interventions developed for children with attachment disorders and severe disturbances (the evidence reviewed predates the DSM-V distinction between reactive attachment disorder and disinhibited social engagement disorder). Only one program was deemed to have "promising research evidence," Dyadic Developmental Psychotherapy (see below). Notably, no program was deemed a ranking of "supported by research evidence," and many of the proposed programs developed specifically with attachment disorder were not able to be rated because of the lack of relevant evidence.

Notwithstanding the very limited evidence of effective treatments, we discuss a range of treatment approaches that have been proposed. One set of studies from uncontrolled trials uses behaviorally based approaches for attachment disorder. Mukaddes and colleagues (Mukaddes, Bilge, Alyanak, & Kora, 2000; Mukaddes, Kaynak, Kinali, Besikci, & Issever, 2004) reasoned that PDD, autism, and RAD/DSED share similar symptoms and so may all benefit from comparable treatment approaches. The first study included 15 children (nine boys, six girls) diagnosed with RAD who had initially been misdiagnosed with PDD. These children had a history of pathogenic care and showed impairments in social interaction, communication, and language development. The treatment involved participation with their current primary caregivers in the 3-month TEACCH outpatient program. Following treatment, children showed improvements in social behavior (increased eye contact, reciprocal play, and social imitation) and ability to form sentences and decreased stereotyped behavior. In a second study, Mukaddes et al. (Mukaddes et al., 2004) used a home-based version of TEACCH and compared the progress of 10 boys with diagnoses of autism to 11 boys and girls with diagnoses of RAD, all of preschool age. Children in both the autism and the RAD group showed overall improvements, and children with RAD showed greater improvement than children with autism on the total development score on the subscales of language-cognition, social and self-care, and fine and gross motor skills.

Becker-Weidman (Becker-Weidman, 2008) examined the effectiveness of DDP, an intervention described above that was developed by Hughes (Hughes, 2004). Following treatment, caregivers of children in the DDP group reported significant decreases in problem behavior and symptoms of attachment disorder. Studies of DDP provide an interesting start to developing an evidence base, but, as noted in the CEBC, the evidence base is not now sufficient, and there remains too little evidence using rigorous randomized controlled trial designs.

One of the more ambitious treatments for RAD-related problems was carried out in an institutional setting in Romania (Smyke et al., 2002). Smyke and colleagues assessed three groups of children. One group consisted of 32 children, aged 4–68 months, who were living in a conventional institutional setting with approximately 30–35 children looked after by two or three caregivers; a second group of children (N = 29; aged 18–70 months) were living in an experimental institutionalized program that reduced the child-caregiver ratio and reduced the number of caregivers with whom the children had regular contact; the third group (N = 33) of children included never-institutionalized children. Results indicated that children in the conventional institutional setting exhibited substantially more inhibited and indiscriminate behavior than children in the pilot institutional setting and those in the noninstitutionalized group. This study demonstrated the strongest reduction in RAD-like behaviors so far reported – although the comparison group is composed of children who remained in institutional care.

Foster care is an index of the kind of severe or "pathogenic care" that would constitute a risk for attachment disorder. Accordingly, interventions for children in foster care may yield valuable leads. A Cochrane Review (Turner, Macdonald, & Dennis, 2007) noted that there is "no evidence that training foster carers in cognitive-behavioral methods has a significant impact on psychological functioning in looked after children" (p. 15); neither was there evidence for a change in placement breakdown, a particularly important outcome for attachment disorder

behavior. A separate Cochrane Review of children in treatment foster care (Macdonald & Turner, 2008) found more positive evidence for reducing behavioral/emotional problems; however, attachment disorder behavior was not specifically targeted. As a result, any relevance to attachment disorder can only be indirect.

Several other specific studies warrant mention. An intervention trial for foster parents conducted by Minnis et al. (Minnis, Pelosi, Knapp, & Dunn, 2001) is one of few to assess attachment disorder behaviors alongside more typical behavioral/emotional problems in the child. The 3-day training program, which was focused on communication and attachment, did not significantly improve child behavioral/emotional problems, including RAD-related behavioral problems. Dozier and colleagues have designed an intervention for infants and toddlers in foster care, "Attachment and Biobehavioral Catch-up" (Dozier, Peloso, Lewis, Laurenceau, & Levine, 2008; Dozier, Stovall, Albus, & Bates, 2001; Stovall-McClough & Dozier, 2004; Stovall & Dozier, 2000). In several studies, they describe their intervention for children and their foster caregivers to address, among other factors, the caregiver's ability to respond to the challenging and perhaps unusual signals of the young child. The attachment-based intervention consists of ten 60-min sessions in caregivers' homes. Effectiveness reports are still somewhat preliminary, but it seems to be the leading intervention model for infants in the foster care system. An additional set of studies for somewhat older children in foster care was reported by Fisher and colleagues (Fisher, Gunnar, Chamberlain, & Reid, 2000) using a cognitive-behavior and social learning model. Results are also somewhat preliminary; the promise for providing data that may be at least indirectly relevant to managing attachment disorder symptoms is considerable.

Finally, although adoption is rarely seen as a "treatment," there is no intervention that is more substantial and pervasive. Studies of children adopted following early institutional rearing provide the relevant information about expectable outcomes of children with attachment disorder. Several lessons are worth reporting from existing studies. Most importantly, as noted above, a sizable minority of children show persisting attachment disorder behavior years after adoption, offering a caution about the amount of gain that might be made with a brief treatment.

Clinical Case Example

Patient B was 4 years old when she was first referred for psychiatric services. At her initial visit, she reached up for high fives from the front desk staff. While her vital signs were collected, she darted into two different offices, climbing onto the laps of therapists and smiling as she banged on their keyboards. Her adoptive mother, a kind and capable woman, smiled with a mix of frustration and affection as she attempted without success to corral B.

B had been removed from her biological parents at 30 months. She had experienced profound neglect and sexual abuse. She had been born full term, without perinatal exposure to drugs or alcohol and had an average birth weight and size. She was briefly hospitalized after birth with RSV. However, when she was removed from care 2 years later, she was underweight and had marked physical and verbal developmental delays and many concerning behaviors. She was briefly placed with a temporary foster family before transfer to the foster family that would ultimately adopt her.

B's adoptive parents were married and from generally middle-class backgrounds. B's adoptive mother took time off from working in order to stay home with her until B entered full-time schooling. Under their care, B became well nourished and her physical delays normalized. However, her speech remained markedly delayed, particularly social or pragmatic speech, and her behavior was significant for difficulty with sleep, night terrors, hyperactive behavior, impulsivity, and fidgeting. Most significant to B's adoptive parents was their difficulty developing an "intimate" relationship with her. They described her as happy and caring but seemingly ambivalent about where she received support and without a sense of "stranger danger."

B was evaluated by several different kinds of clinical professionals; her behavior was something of a mystery to these professionals and her parents. She was evaluated by a developmental pediatrician because her speech delays and fidgeting behaviors had raised flags for autistic spectrum disorder. Extensive evaluation and follow-up definitively ruled out ASD because she had clear social reciprocity and extensive pretend play. At this point, she entered preschool and was referred for special education services, occupational therapy, and speech therapy. Psychoeducational testing indicated an IQ in the low-average range, and an IEP was implemented. She also was assessed at an advocacy center by a therapist that specialized in childhood abuse. While B's play reflected themes of her past abuse and her nighttime terrors suggested trauma associated nightmares, post-traumatic stress disorder did not fully explain her hyperactivity and superficial engagements. She was also diagnosed with attention deficit hyperactivity disorder by her pediatrician, had clear elevations on screening measures, and was treated with stimulants which resulted in partial positive response. In both school and at home, B continued to have inappropriate social boundaries, running away behaviors, and impulsive conversation with strangers.

In the psychiatric clinic, B was evaluated with an individual interview, parent interviews, parent-child observations, and screening tools. Collateral information was collected from other providers and from school, and ongoing assessments were made as B engaged in individual and dyadic therapy and her adoptive mother engaged in parent education and counseling. B's constellation of symptoms was understood as an attachment disorder with a disinhibited style of social engagement. Her ongoing therapy focuses on development of the relationship between B and her adoptive mother, behavior modification in order to ensure her safety, ongoing pragmatic language work in order to facilitate mentalization, and medical treatment with methylphenidate and clonidine for comorbid disorders. Treatment gains have so far been notable for reductions in symptoms of ADHD, with some improvements according to mother report in B seeking out her parents when distressed. This case highlights the rare but significant presentation of marked symptom impairment that resulted from noninstitutional early childhood neglect, the importance of collaboration with multiple different systems of care, and the complication of comorbid psychiatric disorders that may also require targeted treatment.

Conclusion

The current evidence base on attachment disorders is substantial compared to what was available for previous diagnostic iterations. This chapter highlighted some of the particular advances, including a substantial increase in clinical research papers on the social, cognitive, neuroscience, and other biological correlates of attachment disorder behaviors; a consideration of attachment disorder and related behaviors from many countries in a broader array of populations, beyond the population of children who experienced institutional deprivation; and a collection of studies using diverse research and clinical measures, including but not limited to those derived from attachment research.

On the other hand, clinical research evidence on treatment remains inadequate for providing clear and significant direction for managing or reducing the key features of attachment disorder in young and older children and its effects on the family and the child's social and scholastic settings. Also uncertain is the basis for clinical decision-making concerning the co-occurring symptoms routinely found in children with an attachment disorder. There is some suggestion that many of the co-occurring disorders found in children with an attachment disorder warrant their own targeted treatment (e.g., stimulants for ADHD), although the evidence base for that is essentially anecdotal and in any event quite limited. It is possible that treatment gains for these co-occurring disorders may be different in children with an attachment disorder, perhaps because the causal mechanisms may differ from what is typically found or because the symptoms of co-occurring conditions are confounded when

linked with attachment disorder. Further anticipated refinement of the diagnostic features of RAD/DSED may benefit from the current waves of study, but it is in the area of clarifying treatment options that clinical research now needs to focus.

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Disinhibited Social Engagement Disorder

14

Devi Miron and Charles H. Zeanah

This chapter provides an overview of a disorder newly defined in DSM-5 (American Psychiatric Association [APA], 2013), disinhibited social engagement disorder (DSED). This disorder has been recognized and described by various names since at least the mid-twentieth century and defines a pattern of behavior characterized by the absence of developmentally expected reticence in young children about engaging and interacting socially with unfamiliar adults. These defining behaviors have been identified most readily in young children raised in institutions or in those who have been neglected and placed in foster care.

Although the behaviors comprising the disorder have been known for some time, the term DSED was introduced in DSM-5 (APA, 2013). We begin by providing a brief history of the development of this diagnosis in the *Diagnostic and Statistical Manual of Mental Disorders* (DSM). Although to date no studies have been conducted formally assessing the reliability and validity of the new DSM-5 (APA, 2013) DSED criteria per se, reviews have documented a large number of studies that form an empirical justification for the diagnosis of DSED (see Zeanah & Gleason, 2015). In this chapter we update and expand on these reviews, and we consider the symptom presentation, etiology of the disorder, associated impairments and comorbidities, course and developmental challenges, intervention, and what the data suggest regarding prognosis and outcome. In addition, we consider whether it is appropriate to consider DSED as an attachment disorder. We conclude with recommendations for further study.

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History of DSED in the DSM

Attachment disorders were first mentioned in official psychiatric nomenclatures under the diagnosis of "Reactive Attachment Disorder (RAD)" in 1980 in the third edition of the DSM (DSM-III) (APA, 1980). In this edition, RAD was equated with failure to thrive, and the diagnosis specified that the onset occurs before the age of 8 months. Given that the typical age of selective attachment formation is between 7 and 9 months, infants were required to develop the disorder before actually forming a selective attachment. The criteria were substantially revised in DSM-III-R (APA, 1987). Here, DSM dropped the failure to thrive requirement and changed the onset to within the first 5 years of life. It was also in the DSM III-R that two types, "inhibited" and "disinhibited" RAD, were introduced. These types persisted into the publication

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of the DSM-IV (APA, 1994), and only minor changes to the criteria were made.

Perhaps the most important early study informing the criteria for contemporary DSED nosology was conducted with 26 young children being raised in residential nurseries in London by Barbara Tizard and her colleagues (Tizard, 1977). Eight of the children were described as emotionally withdrawn and socially unresponsive, and ten others were indiscriminately social, attention seeking, and clingy with everyone, including unfamiliar adults. The remaining eight actually formed selective attachments to caregivers and less often approached strangers (Tizard & Rees, 1975). The two attachment disordered phenotypes in the Tizard study – emotionally withdrawn and indiscriminately social – were later incorporated into criteria in formal nosologies, which defined two basic clinical presentations of disordered attachment in young children and were recently revised as RAD and DSED.

For almost 20 years after appearing in DSM-III, the disorders attracted little attention from investigators, so until DSM-5, revisions to criteria were made largely in the absence of any relevant research (Zeanah, 1996). In fact, the first study directly addressing the validity of any criteria did not appear until 1998 (Boris, Zeanah, Larrieu, Scheeringa, & Heller, 1998). With the publication of DSM-5 came substantial changes to the diagnostic criteria of RAD, including defining the two phenotypes as two distinct disorders rather than subtypes of the same disorder. This was more similar to the approach taken in ICD-10 (World Health Organization [WHO], 1992) in which reactive attachment disorder was distinguished from disinhibited attachment disorder.

In DSM-5, RAD and DSED are classified under trauma- and stressor-related disorders. RAD includes only a description of the "inhibited" attachment presentation, and the DSED has replaced RAD, "disinhibited" type. The rationale for the changes was that other than arising in similar conditions of social neglect, the two disorders differ in most other important ways, including phenotypic characteristics, correlates, course, and response to caregiving or intervention (Rutter et al., 2009; Zeanah & Gleason, 2015). For example, depressive signs are seen in children with RAD, and patterns of impulsivity are more commonly associated with DSED (Gleason et al., 2011). Further, RAD resolves nearly completely with access to an adequate attachment figure, whereas DSED can persist in the context of adequate caregiving and a selective attachment relationship (Zeanah & Gleason, 2015). In light of this, lack of selectivity of attachment figures was one criterion that was eliminated from the DSM-IV RAD description with the publication of DSM-5.

Clinical Presentation

In order to meet criteria for DSED according to the DSM-5, the child must have a developmental age of at least 9 months (APA, 2013) because that is when focused attachment typically develops. Prior to 9 months and the appearance of stranger wariness, socially disinhibited behavior is normative. Core social and behavioral features of DSED include venturing off without checking in with caregivers and lack of wariness of strangers, including inappropriate approach to unfamiliar adults and a willingness to depart with them without a caregiver. Children with DSED also demonstrate a lack of appropriate physical and social boundaries, such as interacting with adult strangers in overly close proximity and by actively seeking close physical contact or comfort. By the preschool years, verbal boundaries may be violated as the child asks overly intrusive and overly familiar questions of unfamiliar adults. These behaviors have been reported in numerous studies and comprise a set of objectively defined signs of disorder (e.g., see Chisholm, 1998; O'Connor & Zeanah, 2003; Tizard & Rees, 1975; Zeanah, Smyke, & Dumitrescu, 2002).

The criteria for DSED in DSM-5 also distinguish socially disinhibited behavior in the disorder from the impulsivity displayed by individuals with ADHD. This is included in the DSM description because several lines of evidence suggest that some signs of ADHD and of DSED overlap. Nevertheless, it is clear that children may have ADHD without socially indiscriminate behavior, or socially indiscriminate behavior without ADHD, but there are often moderate correlations between the two symptom profiles (Gleason et al., 2011).

DSM-5 criteria for DSED further specify that the child must have experienced insufficient care such as social and emotional neglect, repeated changes in primary caregivers, or rearing in unusual settings that limit the opportunity to form stable attachments, such as institutions. This insufficient care is presumed to be responsible for the pattern of indiscriminate social behavior. According to Zeanah and Gleason (2015), tying the phenotype to grossly inadequate caregiving was retained in DSM-5 for the important reason that children with a known biological abnormality do not necessarily qualify for the diagnosis of DSED. For example, children who have Williams syndrome – a chromosome 7 deletion syndrome – have been reported to demonstrate phenotypically similar behavior to those with DSED, even though the children have received adequate care.

Functional Impairment

Some have suggested that indiscriminate behavior represents an adaptive quality in the context of environmental deprivation. Seeking social contact and interaction could signify an effort to compensate for lack of expectable responsiveness. Chisholm (1998), in her longitudinal study of children adopted into Canada from Romanian institutions, reported that adoptive parents did not view their children's indiscriminate behavior as a problem. On the other hand, Gleason et al. (2011) showed that indiscriminate behavior was associated with functional impairment concurrently and predictively. Many symptoms may arise as efforts at adaptation, though the evidence to date suggests these behaviors are maladaptive.

Quite aside from the physical dangers of a child who does not stay close in unfamiliar settings and with strangers, there are other concerns about indiscriminate behavior. For example, a relatively unexplored hypothesis is that the behaviors are associated with abnormalities in social cognition – in this case recognizing and responding adaptively to social boundaries – that portend later interpersonal difficulties for the child. In one of the few longitudinal studies to follow children with signs of DSED into adolescence, for example, Hodges and Tizard (1989) reported serious peer relational difficulties.

Etiology

Many factors have been suggested as potential risks for DSED, including biological factors, cognitive factors, subnutrition, attachment status, dose of deprivation, and genetic factors (Zeanah & Gleason, 2015). Insufficient caregiving appears to be necessary but not sufficient to cause the disorder because most children raised in conditions of deprivation do not display manifestations of DSED. Preliminary examination of the specific features of social neglect/deprivation experienced by children who develop DSED as well as child risk factors has been demonstrated, though better characterization of intrinsic and extrinsic risk factors is needed.

Neglectful Caregiving Environments

Children who have experienced seriously adverse, neglectful caregiving environments have demonstrated clear increased risk for indiscriminate social behavior compared to children who have not been exposed to adverse conditions (Boris et al., 2004; Bruce, Tarullo, & Gunnar, 2009; Chisholm, 1998; Gleason et al., 2011; O'Connor & Rutter, 2000; Oosterman & Schuengel, 2007; Pears, Bruce, Fisher, & Kim, 2010; Smyke, Dumitrescu, & Zeanah, 2002; Van Den Dries, Juffer, van Ijzendoorn, Bakermans-Kranenburg, & Alink, 2012; Zeanah et al., 2004; Zeanah, Smyke, Koga, Carlson, & the BEIP Core Group, 2005). These environments can exist in child welfare institutions (so-called congregate care) as well as in neglectful conditions leading to intervention by child protective services (e.g., foster care). Below, we review the relative contributions of the environments to the development of DSED.

Pre-institutional Factors In a study of institutionalized young children in Portugal, Oliveira et al. (2012) found that maternal prenatal risk,

operationalized as having a physical disease, abusing substances, limited prenatal care, and preterm birth, predicted indiscriminate behavior, as did emotional neglect. They also demonstrated that neglect mediated the association between maternal prenatal risk and indiscriminate behavior. On the other hand, Gleason et al. (2014) found that institutionalized children's prenatal environment was not related to indiscriminate behavior.

Institutional Factors Signs of DSED have been attributed to being raised in an institution by multiple, rotating caregivers (Dobrova-Krol, Bakermans-Kranenburg, van IJzendoorn, & Juffer, 2010; Gleason et al., 2014; Smyke et al., 2002; Zeanah et al., 2005) and/or without a preferred caregiver (Soares et al., 2014). For example, in the first study conducted on disturbances of attachment in children living in an institution since the Tizard study, Smyke et al. (2002) demonstrated that signs of indiscriminate social behavior as observed in DSED were associated in stepwise fashion with increasingly adverse caregiving environments, at least as indexed by caregiver-child ratios. Further, they found higher rates of indiscriminate behavior among all institutionalized children regardless of the number of caregivers compared with children raised in families attending childcare centers. The results indicated that limited opportunities to form attachments in the caregiving environments were associated with increasing risks for DSED.

In addition to number of caregivers, caregiving quality in institutions has been studied as a risk factor for the development of DSED (Dobrova-Krol et al., 2010; Gleason et al., 2014; Zeanah et al., 2005). However, the findings are mixed.

To attempt to determine which components of caregiving quality are specifically associated with the risk of development of DSED or RAD, Zeanah et al. (2005) examined naturalistic interactions between institutional caregivers and young children, ages 12–31 months in the Bucharest Early Intervention Project (BEIP), a randomized controlled trial of foster care as an alternative to institutional care (Zeanah et al., 2003). They found that institutionalized children displayed more signs of disinhibited RAD than never institutionalized children in the community. Further, for the institutionalized children, a composite variable of caregiving quality was associated with formation and organization of attachment and signs of emotionally withdrawn/inhibited RAD but was unrelated to indiscriminate behavior.

Gleason et al. (2014) found that including children from all groups in the BEIP (institutionalized, previously institutionalized foster care, and never institutionalized), lower quality of caregiving contributed to increased indiscriminate social behavior at 54 months. In children with histories of institutional rearing, neither caregiving quality at baseline nor percent time in institutions was associated with indiscriminate behavior. The authors suggested that the variations in institutional care quality or duration in the first 17 months of life were not, in of themselves, determining factors of indiscriminate behavior.

In their study of HIV-infected young children raised in institutions and families, however, Dobrova-Krol et al. (2010) found higher levels of positive caregiving among institution-reared children with indiscriminate social behavior, an association in the opposite direction as that seen in home-reared controls. The authors concluded that indiscriminate behavior might serve a different function for children reared in institutions and suggest that the association between indiscriminate behavior and caregiving quality in institutions may reflect a "natural reaction of caregivers to contact-inducing behaviors" (p. 1374).

More research is needed, but it is possible that inadequate care (as often occurs in institutions) may lead to different problems at different thresholds, that is, more severe deprivation leading to signs of RAD and less severe leading to signs of DSED in vulnerable children.

Noninstitutional Caregiving Environments Lyons-Ruth, Bureau, Riley, and Atlas-Corbett (2009) found that indiscriminate behavior was present in high-risk, family-reared 18-month-old infants only if they had been maltreated or if their mothers had a history of psychiatric hospitalizations. Further, quality of interaction, or maternal disrupted communication, specifically

disorientation (e.g., appearing frightened of the infant, disoriented wandering), mediated the relationship between the caregiving risk and indiscriminate behavior. In fact, the researchers observed that these mothers displayed uncomfortable and awkward patterns of interacting with their infants, and they likened this to caregiver interaction in understaffed institutions. They suggest that indiscriminate social behavior may be related to the depth of emotional engagement of caregivers rather than to physical neglect or rotating caregivers, per se.

Pears et al. (2010), studying children in foster care, found signs of DSED were related to the number of placement disruptions rather than severity of maltreatment (defined as physical neglect, supervisory neglect, and emotional maltreatment). Foster children who had greater numbers of foster caregivers demonstrated poorer inhibitory control and that this (but not severity of emotional maltreatment) was related to indiscriminate social behavior. Based on these results, the authors concluded that indiscriminate behavior is part of a larger pattern of dysregulation, which is associated with inconsistency in caregiving. This finding supports the inclusion in DSM-5 of repeated changes in caregivers as a type of insufficient care required for the diagnosis of DSED.

Child Vulnerability Factors

Sensitive Period with Regard to Adequate Caregiving Several researchers have suggested a sensitive period exists with regard to adequate caregiving and the display of disinhibited social behavior (e.g., Dobrova-Krol et al., 2010; Rutter et al., 2007). In the English and Romanian Adoptees Study (ERAS), researchers found evidence of a sensitive period. That is, 27 of 29 children who showed persistence of indiscriminate behavior through 15 years were adopted *after* 6 months of age (Rutter et al., 2010). Smyke et al. (2012) also reported that children removed from institutional settings and placed in foster care prior to 24 months were significantly less likely to show signs of indiscriminate behavior through 54 months of age. Although additional research on this question is needed, it appears that the earlier the child is removed from adversity, the less likely that she/he will show signs of indiscriminate behavior.

Biological Factors and Subnutrition Results are mixed about associations of growth delays and indiscriminate behavior. Although Johnson, Bruce, Tarullo, and Gunnar (2011) found that only post-institutionalized children with stunted growth at adoption were at risk for indiscriminate behavior, it is hard to know if the growth stunting itself is implicated or if stunted growth merely indexes more severe deprivation and that is what led to indiscriminate behavior. In the ERAS, for example, there was no clear association between small head circumference or subnutrition and disinhibited attachment, although they noted that head growth partially mediated the association between institutional care and indiscriminate behaviors (Rutter, O'Connor & the English and Romanian Adoptees (ERA) Study Team, 2004; Sonuga-Barke et al., 2008).

Genetic Factors The biological substrate of socially indiscriminate behavior is unknown. However, because not all children who experience early neglect or other adverse caregiving environments develop indiscriminate behavior, biological or genetic factors contributing to the behavior must be considered. While there has been little research to this effect to date, recently a number of investigators have proposed models of gene x environment contributions to indiscriminate social behavior.

Soares, Belsky, Mesquita, Osorio, and Sampaio (2013) proposed that studying children with Williams syndrome might provide insight into the development of indiscriminate social behavior. In their gene x environment model, they propose that children will be most likely to display indiscriminate behavior when they carry specific polymorphisms within the Williams syndrome critical region (the portion of the 7th chromosome the deletion includes) and when they have experienced low quality of care typical of many institutions. This model has not yet been tested.

Bakermans-Kranenberg, Dobrova-Krol, and van IJzendoorn (2011) examined whether the serotonin transporter gene (5HTT) moderated the association between institutional care and attachment security, disorganized attachment, and indiscriminate behavior among young children raised in Ukrainian institutions. Their rationale was that in a number of gene x environment interaction studies, the 5HTT genotype has been shown to moderate the effects of negative environmental influences. The results of the study were that the long allele of the genotype protected against the development of disorganized attachment in children reared in institutions; however, it did not buffer against the development of indiscriminate behavior.

Drury et al. (2012) examined vulnerability to the development of indiscriminate social behavior within the context of BEIP. Specifically, they studied the effect of the interaction between group status (random assignment to foster care vs. remaining in institutional care) and functional polymorphisms in the serotonin transporter gene (5HTT) and in brain-derived neurotrophic factor (BDNF) on levels of indiscriminate behavior over time. They demonstrated that children with the combined short allele, s/s, 5httlpr genotype, and the met66 carriers of BDNF ("plasticity genotypes") demonstrated the lowest levels of indiscriminate behavior in children randomized to foster care but the highest levels of indiscriminate social behavior in children randomized to care as usual. Children with either the long allele of the 5httlpr or val/val genotype of BDNF demonstrated intermediate levels of indiscriminate behaviors over time and no group by genotype interaction. The researchers concluded that individual differences in responsiveness to changes in the caregiving environment are influenced by these specific genetic variations. The findings illustrate a "differential susceptibility" model of gene x environment interactions in children exposed to deprivation (Belsky, Bakermans-Kranenburg, & van IJzendoorn, 2007). The results also suggest why only some children exposed to serious adversity might develop indiscriminate behavior and why only some children may benefit from an improvement in caregiving quality over time.

Disorganized Attachment In BEIP, Gleason et al. (2014) examined a variety of factors at a baseline assessment when 136 children 6–30 months were living in institutions as predictors of indiscriminate behavior at 54 months (when most of the children were no longer living in institutions). They found that the best predictor of indiscriminate behavior at 54 months was disorganized attachment behaviors. Among children with lower levels of disorganization, caregiving quality was associated with lower rates of indiscriminate behavior. The authors suggested that children who have the capacity to develop organized attachments with a preferred caregiver in an institution also might be able to develop more discriminating social behaviors later.

Associated Impairments and Comorbidities

Attachment and Attachment Disorders

One aspect of considering how attachment relates to DSED in part involves considering how classifications and quality of attachment relate to DSED. Among institutionalized children, Soares et al. (2014) found that the absence of a preferred caregiver predicted greater presence of indiscriminate social behaviors even after taking into consideration pre-institutionalization conditions, including prenatal and maternal risk. Other studies have found that even when institutionalized children had preferred caregivers, they displayed significantly higher levels of indiscriminate behaviors compared with family-reared children (Dobrova-Krol et al., 2010; Smyke et al., 2002; Zeanah et al., 2005).

The limited research to date has demonstrated a significantly lower rate of children with a clearly defined attachment pattern compared with never institutionalized children (Dobrova-Krol et al., 2010; Zeanah et al., 2005). In addition, even among children with a clearly defined attachment pattern, there seems to be a higher prevalence of disorganized attachment and a lower prevalence of secure attachment as assessed by categorical and continuous ratings of attachment among

institutionalized children compared with noninstitutionalized children (Dobrova-Krol et al., 2010; Vorria et al., 2003; Zeanah et al., 2005). However, the presence of an organized attachment relationship, ratings of attachment (categorical or continuous), and attachment security have been found to be unrelated to disinhibited behavior in institutionalized children (Dobrova-Krol et al., 2010; Zeanah et al., 2005).

Although quality of caregiving has been shown to predict attachment rating on a continuous scale, there have been mixed results from assessments of the relationship between quality of caregiving and disinhibited social behavior in children living in institutions. Zeanah and colleagues (2005) found no such relationship. Curiously, Dobrova-Krol et al. (2010) found that indiscriminate behavior was associated with *higher* levels of positive caregiving for institutionalized children, but not for family-reared children, suggesting that indiscriminate behavior may serve different functions for children being reared in institutions.

Among children with histories of institutional rearing in the BEIP, Gleason et al. (2011) found that nearly half of children displaying indiscriminate social behavior had organized attachment classifications. However, for toddlers aged 42 months, attachment security was inversely related to indiscriminate behavior although some securely attached children showed signs of DSED. When the researchers dichotomized attachment classifications into typical (secure, avoidant, or ambivalent) or atypical (disorganized, controlling, or insecure-other) , atypical attachment was moderately associated with signs of DSED.

A few studies have revealed indiscriminate social behavior in children with secure attachment relationships (e.g., Chisholm, 1998; O'Connor, Marvin, Rutter, Olrick, & Britner, 2003). In contrast, Van Den Dries et al. (2012) found no relationship between indiscriminate behavior and either a secure or disorganized attachment relationship. Further, Bruce et al. (2009) found that disinhibited social behavior was not significantly correlated with attachment-related behaviors.

The most commonly found attachment classifications in young children with indiscriminate social behavior are disorganized or insecure-other.

In summary, children with DSED may or may not have selective attachments. That is, they may have secure, insecure, disorganized, or no attachments (Bakermans-Kranenberg et al., 2011; O'Connor et al., 2003; Smyke et al., 2002; Zeanah et al., 2005). The fact that clinically significant levels of indiscriminate behavior occurs across an entire range of attachments was a large part of the rationale behind the reconceptualization of DSED in DSM-5. In particular, children having a secure attachment ought to be incompatible with having an attachment disorder. Some have argued, however, that because the disorder arises from the absence or disturbances in the child's original attachment relationship, it makes more sense to consider it a disorder of attachment (Lyons-Ruth, 2015).

Developmental Delays and Behavioral Impairments

Although children with a history of adverse caregiving, particularly institutionalization, often display developmental delays (Nelson et al., 2007), and sometimes signs of autism spectrum disorder (Levin, Zeanah, Fox, & Nelson, 2015; Rutter et al., 2007), on the whole, research findings do not support consistent comorbidity with DSED.

Cognitive Ability Mixed results have been reported with respect to IQ and its coexistence with indiscriminate social behavior. The ERAS revealed no association between mental age at entry into the UK (majority <24 months) and indiscriminate behaviors at 6 years old (Rutter & O'Connor, 2004). Lyons-Ruth et al. (2009) found no relationship between Bayley Mental Development Index scores and socially indiscriminate attachment behavior. Bruce et al. (2009) found no relationship between socially indiscriminate behavior and general cognitive ability.

In contrast, Van Den Dries et al. (2012) found that children with higher cognitive scores showed less indiscriminate behavior. Further, BEIP reported a moderate association between baseline (mean of 22 months) developmental quotient and indiscriminate behaviors at 54 months (Gleason

et al., 2014). However, IQ at age 54 months was not associated with the presence of indiscriminate behavior.

Of course, both cognitive impairment and indiscriminate social behavior are outcomes of deprived environments, but this does not mean that they are causally related. It is difficult to assess whether intellectual disability predisposes to indiscriminate behavior when both result from the same environmental risk. In any case, indiscriminate social behaviors are clearly not solely the result of delays in cognitive development.

Externalizing Problems Given the intrusive social behaviors displayed by children with signs of DSED, several studies have examined externalizing symptoms and behaviors in these children. Among institutionalized toddlers, Zeanah et al. (2002) found no relationships between caregiver reports of indiscriminate behavior and their global ratings of children's aggression. Similarly, in the BEIP, there was no association between indiscriminate and aggressive behaviors in children ages 42 months and younger (Gleason et al., 2011; Zeanah et al., 2005). However, Lyons-Ruth et al. (2009) showed that toddlers who were indiscriminate with strangers showed more aggressive and hyperactive behavior problems in kindergarten.

Signs of DSED were associated with symptoms of ADHD and modestly associated with signs of disruptive behavior disorders for children aged 54 months involved in the BEIP (Gleason et al., 2011). This outcome replicated similar findings in other studies of Romanian adoptees. Chisholm (1998) found that children's scores on a measure of indiscriminate behavior were significantly correlated with externalizing scores on the Child Behavior Checklist. Rutter et al. (2007) found that 35% of children with mild or marked "disinhibited attachment" at age 6 or 11 had challenges with inattention and overactivity at age 11. However, despite these correlations, it is clear from the existing research that ADHD and DSED are distinct clinical entities. For example, in BEIP, only four of the 20 children who met criteria for ADHD also met criteria for DSED, and only four of the 16 children who met criteria for DSED also met criteria for ADHD (Gleason et al., 2011).

Inhibitory Control Given the association between socially disinhibited behaviors and attention, hyperactivity, and impulsivity problems, several studies have investigated the relationship of signs of DSED to inhibitory control and other neurobiological processes (see Zeanah & Gleason, 2015, for a review). Pears et al. (2010) found that inhibitory control negatively associated with indiscriminate friendliness even after controlling for general cognitive ability. Bruce et al. (2009) found that children with poorer inhibitory control were more likely to display indiscriminate social behaviors. In addition, they found inhibitory control mediated the relationship between disinhibited social behavior and length of time in an institution.

Course and Developmental Challenges

In the BEIP signs of DSED were moderately stable from ages 30 months to 54 months for children with a history of institutional rearing (Gleason et al., 2011). Among the continuously institutionalized group, stability was even higher.

For children adopted out of institutions, signs of DSED have shown moderate stability. The previously institutionalized children in the Tizard study continued displaying "over-friendly" and attention-seeking behavior from age 4 to 8 years, even among those who were adopted. The researchers noted that once established, over-friendly behavior was especially resistant to change (Tizard & Hodges, 1978; Tizard & Rees, 1975). Those children adopted out of Romanian institutions into Canada showed increased levels of indiscriminate sociability 2–4 years post adoption (Chisholm, 1998). Further, in the ERAS, signs of indiscriminate behavior in children who were adopted out of institutions prior to the age of 42 months were persistent from 6 to 11 years of age (Rutter et al., 2007).

To date, only two longitudinal studies have tracked the course of DSED behaviors through

adolescence. Rutter et al. (2010) identified a group of 29 children who show continuously elevated signs of DSED from early childhood through 15 years of age. At age 16, in the Tizard study indiscriminate behavior with caregivers was reduced but was evident with peers. Relations with peers were conflicted and superficial, for example, naming a recent acquaintance as a close friend (Hodges & Tizard, 1989). Additional research is needed to specifically monitor the stability of the newly defined DSED over time.

In addition to behavioral and attachment difficulties, studies of children with signs of DSED indicate that the developmental challenges of children with DSED include socioemotional (Gleason et al., 2011), peer relational (Hodges & Tizard, 1989), and educational (Rutter et al., 2007) impairments. Signs of DSED in the BEIP were concurrently associated with lack of social-emotional competence at 30 and 42 months and with functional impairment at 54 months. Lyons-Ruth et al. (2009) found that socially indiscriminate behavior at age 18 months predicted behavior problems, including hostile behavior and hyperactivity at age 5 years. Rutter et al. (2007) reported that children with histories of institutional rearing who showed indiscriminate behavior had impaired peer relationships and higher rates of psychopathology. Further, they noted an increase in use of mental health services and more special education received by these children.

Along these lines, Overbeek, Clasien de Schipper, Lamers-Winkelman, and Schuengel (2014) studied a group intervention designed for children, ages 6–12 years with a history of witnessing interparental violence. They found that children with symptoms of DSED were less likely to show reductions in internalizing symptoms compared with participants without DSED, perhaps because the shallowness of their social engagement may have impeded the children from engaging fully in therapeutic group activities. Thus, having symptoms of DSED may interfere with children's recovery from their adverse experiences, whether or not these experiences were the direct cause of their disorder. Taken together, these findings indicate that DSED is moderately stable and often significantly impairing.

Intervention

To date, there have been few published studies of interventions designed specifically for children with DSED and its associated behaviors. The existing research describes the effects of children displaying symptoms of DSED being removed from institutions and placed with families (e.g., Chisholm, 1998; O'Connor et al., 2003; Smyke et al., 2012; Tizard & Rees, 1975). The "intervention" in these studies is family rearing (foster care or adoption) for children with a history of institutional rearing.

The results have been mixed regarding the effectiveness of adoption for ameliorating symptoms of DSED. For example, Tizard's longitudinal study demonstrated both the persistence of indiscriminate behavior and its reduction following adoption (Hodges & Tizard, 1989; Tizard & Hodges, 1978; Tizard & Rees, 1975). Twenty years later, a longitudinal study of young children adopted into Canada from Romanian institutions revealed significant increases in parent reports of attachment during the first several years following adoption but no comparable decreases in indiscriminate behavior over time (Chisholm, 1998).

Researchers have also attempted to explore which aspects of enhanced caregiving (e.g., sensitivity) are crucial for reducing signs of DSED. In the ERAS, investigators assessed signs of DSED in children adopted into UK families from Romanian institutions at ages 4, 6, 11, and 15 years (O'Connor & Rutter, 2000; O'Connor et al., 2003; Rutter et al., 2007, 2010). They reported little change in the numbers of children with high levels of indiscriminate behaviors between 4 and 6 years, but some decline by age 11 years (O'Connor & Rutter, 2000; Rutter et al., 2007). However, they found no relationship between quality of care in adoptive homes and indiscriminate behavior. This suggests that for

some children, more than enhanced caregiving is necessary to remediate DSED.

In the BEIP, a modest but statistically significant decline in signs of DSED with the foster care intervention was found, although rates of DSED behaviors were significantly lower in the never institutionalized group (Smyke et al., 2012). Notably, within the foster care group, placement before 24 months of age predicted the lowest level of DSED, compatible with a timing of intervention effect.

Given the current knowledge base, it seems prudent to intervene by providing the same caregiving for children with DSED that is known to lead to secure attachment formation – that is, sensitive and responsive to the child's needs. However, existing research suggests that only some children with DSED respond to enhanced caregiving and to varying degrees. Some researchers have proposed that incomplete remediation for those with DSED could reflect individual differences in responsiveness to caregiving (e.g., Drury et al., 2012) or incomplete remediation in those who were most severely affected initially. Further, which aspects of the caregiving environment that need to be enhanced is an area ripe for exploration. Soares et al. (2014) suggested that institutional caregivers be provided training to promote warm, sensitive, and responsive interactions with the child within a context of routines and activities that promote the social-emotional development of the child. Also, Garvin, Tarullo, Van Ryzin, and Gunnar (2012) noted the role that adoptive parents' training preparing them for the types of behaviors their children may exhibit may have played in their findings of the fewer number of formerly institutionalized children with disinhibited symptoms as compared with previous studies. Thus, specific training on minimizing and addressing DSED behaviors could be provided for foster caregivers, adoptive parents, and institutional caregivers alike. Controlled studies are much needed to evaluate these assertions.

Just as adverse care is not in itself sufficient for the development of DSED, interventions must target other contributors to the disorder. Zeanah and Gleason (2015) suggest that evidence-based interventions for which effectiveness has been demonstrated with maltreated children should be attempted with children with DSED (Bernard et al., 2012; Cicchetti, Rogosch, & Toth, 2006), as well as interventions that target social cognitive abnormalities.

Prognosis

Based on the existing research, it seems that young children who develop DSED may or may not respond to enhanced caregiving alone. Preliminary evidence suggests that earlier intervention may be more effective, though the evidence base is quite thin. It is clear that in some cases, signs of the disorder may persist through childhood and into adolescence. DSED has been studied most systematically in younger children. The course of the disorder, including possible changes in symptomatology in middle childhood and adolescence, needs further investigation. Longitudinal, follow-up studies of the sequelae of these disorders in later childhood, adolescence, or adulthood are needed, including peer relationships, interpersonal competence, and other relevant outcomes.

Conclusions

The DSM-5 conceptualization of DSED is congruent with extant research, though much remains to be learned about the disorder. It remains unclear why only some children raised in conditions of deprivation develop signs of DSED. In a similar manner, the specific components of deprivation and putative biological vulnerabilities are not certain. The validity and reliability of DSED criteria also need careful study. Additional research is needed to elucidate the course of the disorder and factors that moderate it. Finally, preventive efforts that specifically target the known causes of the disorder and interventions that address its core features need to be developed and systematically studied.

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Posttraumatic Stress Disorder and Acute Stress Disorder in Childhood and Adolescence

15

Julian D. Ford and Carolyn A. Greene

Posttraumatic stress disorder (PTSD) is a prevalent disorder among children (Copeland, Keeler, Angold, & Costello, 2007) and adolescents (McLaughlin et al., 2013) that can cause clinically significant functional impairment, especially when it occurs with common comorbidities such as anxiety, mood, disruptive behavior, or substance use disorders (D'Andrea, Ford, Stolbach, Spinazzola, & van der Kolk, 2012). Traumatized children also may develop an acute stress disorder (ASD) when traumatic stress symptoms are severe and persistent in the first several days and weeks in the aftermath of exposure to traumatic stressors. In this chapter, we will describe the history of childhood PTSD's origin and changing definitions as it evolved as a theoretical and empirical construct and clinical diagnosis, with a detailed discussion of how PTSD and ASD are defined in the *DSM*-5 (including adaptations for children and separate criteria for diagnosing PTSD in preschool-age children). The chapter then provides a review of empirical evidence regarding epidemiology (prevalence and comorbidities), etiology (risk factors and causal mechanisms, including ethnocultural, gender, and developmental factors), and *DSM*-5 informed assessment, treatment, and prevention of pediatric PTSD.

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History and Definition of Traumatic Stress Disorder Diagnoses

The serious long-term consequences of trauma have been known for centuries (Ray, 2008), but the term PTSD was not officially introduced into the American psychiatric nomenclature until 1980, in the American Psychiatric Association (APA) *Diagnostic and Statistical Manual of Mental Disorders*, Third Edition (DSM-III; American Psychiatric Association [APA], 1980). Prior to this, both the American and the international World Health Organization's diagnostic systems had grappled with how best to classify, describe, and define stress-related diagnoses. In fact, these debates continue into the present day and will be reviewed briefly here.

The first edition of the *DSM*, published in 1952 (APA, 1952), included a gross stress reaction (GSR) diagnosis, which was defined as a "transient situational personality disorder" that could occur when essentially "normal" individuals experienced "severe physical demands or extreme emotional stress." However, GSR was dropped from the second edition of the *DSM* (APA, 1968) and subsumed within the category of adjustment reactions (acute responses to environmental stress of any severity), thus diminishing the diagnosis' specificity (Brett, 1996).

Twelve years later, with the publication of the *DSM-III* (APA, 1980), the extreme emotional

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stress of the GSR resurfaced in an altered form as PTSD, this time with the recognition of the chronic reactions that can occur in response to trauma and differentiation between adjustment and traumatic stress disorders (Brett, 1996). Advocacy efforts by and on behalf of Vietnam War veterans (Frueh, 2007) and women who experienced "rape trauma syndrome" (Burgess & Holmstrom, 1974) were decisive in both restoring a stress-related diagnosis to the formal psychiatric classification system and linking it specifically to the experience of exposure to extreme or traumatic stressors (Breslau & Kessler, 2001).

Further, in an effort to provide a more valid and reliable guide to diagnosis, the *DSM-III* introduced a phenomenological approach to diagnosis and as such provided specific symptom criteria for the diagnosis of PTSD. Exposure to a traumatic stressor (Criterion A) was defined as: "Existence of a recognizable stressor that would evoke significant symptoms of distress in almost everyone" (APA, 1980, p.238). Colloquially, this has been described as a stressor that is outside the range of normal human experience, a distinction that was solidified in the *DSM-III-R* revision(APA, 1987). Twelve possible PTSD symptoms were described, based on early research and clinical observations that identified unwanted and severely distressing memories of traumatic events (technically designated as "intrusive re-experiencing") and extreme states of physiological and emotional arousal (technically designated as "hyperarousal") as core symptoms of PTSD (Horowitz & Smit, 2008).

In a 1994 revision, the *DSM-IV* added subjective features (a response of "intense fear, helplessness or horror") to the traumatic exposure (Criterion A2) and expanded the number of PTSD symptoms to 17, adding attempts to avoid reminders of trauma memories and emotional dysphoria and numbing as a third subgroup (Criterion C). Also added was a Criterion E that specified that the duration of the disturbance had to exceed 1 month. A new traumatic stress-related diagnosis also was added in the *DSM-IV*: acute stress disorder (ASD). Like PTSD, ASD requires a Criterion A traumatic event and includes similar symptom criteria to those constituting PTSD (Criteria B through D). However, ASD cannot be diagnosed unless symptoms and associated impairment last at least 2 days (so as to exclude those with immediate but transient "peritraumatic" reactions, which are almost universal) but not more than 4 weeks following exposure to the traumatic stressor. ASD also includes symptoms of dissociation (i.e., derealization, depersonalization) which were not included in PTSD until the next *DSM* revision (*DSM*-5, see below). ASD is derived from research demonstrating that a distinct subgroup of approximately 10% of individuals experience severely impairing traumatic stress reactions for more than a few days immediately after exposure to traumatic stressors (Bryant, Creamer, O'Donnell, Silove, & McFarlane, 2012; Hansen, Lasgaard, & Elklit, 2013). Adults (Bryant et al., 2012; Shevlin, Hyland, & Elklit, 2014) and children (Bryant, Salmon, Sinclair, & Davidson, 2007) who develop ASD are at high risk for developing PTSD and other psychiatric disorders subsequently, particularly if their ASD symptoms include severe hyperarousal.

During the five decades of evolution of the PTSD diagnosis in the American *DSM* diagnostic system, there has been a parallel process of refinement in the definition of PTSD in the World Health Organization's *International Classification of Diseases* (*ICD*), with several important points of difference. As with the *DSM*, the earliest references in the *ICD* to stress-related responses described transient rather than chronic stress reactions (World Health Organization, 1948, 1955, 1969). The first reference to PTSD appeared in the *ICD-9* (World Health Organization, 1977) under the heading of Adult Onset Adjustment Reaction, which described responses to chronic stress. A separate category of "acute reaction to stress" also was set forth in the *ICD-9* to describe transient responses to "exceptional stress," including separate subcategories reflecting reactions characterized by disturbance of emotions, consciousness, or psychomotor functions (Turnbull, 1998). Thus, while the *DSM-III* did not distinguish acute and chronic posttraumatic stress reactions (categoriz-

ing them all as PTSD) and *DSM-III-R* provided no diagnosis for traumatic stress reactions in the first 30 days after a traumatic stressor, in describing acute reaction to stress as severe symptoms in the immediate wake of experiencing a traumatic stressor, the *ICD-9* was more precise and complete than its *DSM* counterparts.

An updated description of PTSD and acute stress reaction was provided in the most recent (ICD-10; World Health Organization, 1992) edition which was published at approximately the same time that the *DSM-IV* (APA, 1994) was being finalized (Lasiuk & Hegadoren, 2006). The *ICD-10* and *DSM*-IV PTSD and acute stress reaction (*ICD-10*) or disorder (*DSM-IV*) diagnoses involved very similar but not exactly identical definitions of the stressor criterion, symptom criteria, and duration and functional impairment criteria. However, there are several important differences between the *ICD-10* and the *DSM-IV*, some of which foreshadow changes in the *DSM* that occurred 12 years later in the *DSM-5*.

While the DSM-IV continued to describe both PTSD and acute stress reaction diagnoses as Anxiety Disorders (as did the *DSM-III* and *III-R*), the *ICD-10* placed these diagnoses in the "Reactions to Severe Stress and Adjustment Disorders" category. In addition, while the acute stress reaction diagnosis in the *ICD-10* involves a variety of possible symptoms that tend to ebb and flow rather than remaining constant, in the immediate (i.e., up to 3 days) wake of a traumatic experience, the *DSM-IV* requires a more prescribed set of required symptoms, despite evidence that no single set of symptoms characterizes most or all persons who are adversely affected in the acute aftermath of traumatic stressors (Marshall, Spitzer, & Liebowitz, 1999). Finally, an entirely new Disorder of Adult Personality and Behaviour was added to the *ICD-10*, *"enduring personality change after catastrophic experience."* EPCACE must be chronic (i.e., lasting for at least 2 years) and includes some similar symptoms to PTSD (such as emotional numbing, avoidance, and hyperarousal) but is a more chronic existential syndrome that parallels Herman's (1992) complex PTSD. It also anticipates the more complex posttraumatic stress symptoms of emotional and behavioral dysregulation that emerge in an expanded *DSM*-5 conceptualization of PTSD (see below) and that are under review for inclusion as a complex PTSD diagnosis in the forthcoming *ICD*-11 (Cloitre, Garvert, Brewin, Bryant, & Maercker, 2013).

PTSD Diagnostic Criteria Currently in the *DSM*-5 and Proposed for the *ICD*-11

The types of events that qualify as traumatic stressors has been expanded in the American Psychiatric Association's (2013) *DSM-5* to include actual or threatened physical and sexual assaults, abuse, or injury, witnessing severe violence or death (or learning about this occurring to a close family member or friend), or "experiencing repeated or extreme exposure to aversive details of [these types of] traumatic event(s) (e.g., first responders collecting human remains; police officers repeatedly exposed to details of child abuse)" (p. 271). Further, the *DSM*-5 eliminated the requirement that a traumatic exposure must be accompanied by a reaction of fear, helplessness, or horror (A2) based on research showing that almost all individuals who meet the symptom criteria for PTSD also recall experiencing severe peritraumatic distress. It is impossible to discern clinically or scientifically whether memories of past reactions are accurate or partially or fully a byproduct of the current PTSD symptoms (Brewin, Lanius, Novac, Schnyder, & Galea, 2009; O'Donnell, Creamer, McFarlane, Silove, & Bryant, 2010). Importantly, the subjective emotional impact of experiencing traumatic stressors was not lost in the *DSM*-5 PTSD operationalization because it is captured by newly added symptom items.

With regard to symptoms, the *DSM-5* divides PTSD symptoms into four categories, instead of the three categories included in earlier versions (APA, 2013). A number of symptoms of emotional numbing that were grouped together with avoidance in the *DSM-IV* (i.e., inability to feel/express positive emotions, amnesia about important parts of traumatic event(s), feeling

detached from relationships, believing one's life will be cut short—a sense of a foreshortened future) are now placed with new symptoms under the rubric of a new Criterion D. The newly added symptoms reflect negative changes in beliefs and emotions that began during, or worsened after, the traumatic events and include: persistent negative beliefs about oneself, distorted blame of self or others for the traumatic events, and emotional distress in the form of anger, guilt, shame, or horror. Thus, the added symptoms represent trauma-related beliefs and emotional dysregulation that extend beyond fear and anxiety, and, hence, PTSD is no longer grouped in the *DSM*-5 with the anxiety disorders as it was in the *DSM-III* and *DSM*-IV, but in a new class of "trauma and stressor-related disorders" (APA, 2013, p. 271).

With these new negative mood/cognition symptoms, the *DSM-5* PTSD diagnosis now encompasses the trauma-related impairments in self-regulation that hundreds of research studies have shown to be after-effects of exposure to interpersonal traumatic stressors in childhood (e.g., abuse, violent or sexual victimization, family violence) and in some cases in adulthood as well (e.g., genocide, torture, intimate partner violence) (D'Andrea et al., 2012). This is relevant to understanding how and why traumatized youth are at risk for substance use disorders, because the new PTSD dysregulation symptoms are known to be risk factors for and contributors to adolescents' substance use problems (Ford, Hartman, Hawke, & Chapman, 2008).

PTSD hyperarousal symptoms (now Criterion E for the diagnosis) include the *DSM-III* and *DSM-IV* symptoms of severe sleep difficulties, anger and irritability, concentration problems, scanning the environment for potential threat (hypervigilance), and exaggerated startle responses. One prior hyperarousal symptom has been expanded in *DSM*-5 (i.e., posttraumatic anger problems now may include verbally or physically aggressive behavior). Further, an entirely new hyperarousal symptom, reckless or self-destructive behavior, has been added.

In the proposed revisions to the *ICD*-11, PTSD has been simultaneously simplified and expanded. PTSD's three core criteria (re-experiencing, avoidance, and hypervigilance) are to be assessed with only two symptoms each. The expansion would include an alternative complex PTSD diagnosis which encompasses these three core features, as well as persistent and pervasive disturbances in affective, self, and relational functioning that also will be assessed with only two core symptoms each (Maercker et al., 2013).

The *DSM*-5 (APA, 2013) ASD diagnosis has the identical Criterion A (traumatic stressor) with that for PTSD, but differs from "acute" PTSD in several important ways. First, ASD must occur within 1 month of exposure to the stressor event(s) and may be diagnosed as soon as 3 days afterward (in the *DSM-IV* this could be as soon as 2 days after traumatic stressor exposure). Another important difference between ASD and PTSD is that ASD includes fewer symptoms (14 in the *DSM-5*) than PTSD (22, in the *DSM-5*, including the two optional dissociative symptoms). However, every ASD symptom is also a PTSD symptom—or a combination of several PTSD symptoms. For example, in the *DSM-5* the two PTSD intrusive re-experiencing symptoms involving intense or prolonged psychological distress or marked physiological reactions in response to reminders of traumatic events are collapsed into a single ASD symptom. The two optional PTSD dissociative symptoms of depersonalization and derealization also are combined into a single ASD symptom. Several PTSD symptoms that typically do not develop until more than a month following exposure to traumatic stressors are not included in ASD in the *DSM-5*: persistent negative beliefs, blame of self or others, negative emotion states, loss of interest (anhedonia), and feelings of detachment from others.

The most controversial feature of ASD in the *DSM*-IV—which was dropped in the *DSM-5*—was the requirement that at least three of five symptoms of pathological dissociation had to occur either during or after experiencing the stressor event(s). Dissociation is common during and soon after exposure to psychological trauma—this is called "peri-traumatic dissociation" because "peri" signifies that dissociation is occurring close in time to the stressor event(s).

Although peri-traumatic dissociation is a common acute traumatic stress reaction, research has shown that it is neither the only nor the most important such reaction in determining or predicting who will develop PTSD (Bryant et al., 2011). Research also points to three key forms of peri-traumatic dissociation as most prevalent and predictive of adverse outcomes, each of which involves more than a transient feeling of shock or confusion. These do not include feeling emotionally numbed or detached, or "in a daze," which are relatively expectable and immediate reactions of psychological shock. Instead they include feeling a sense of unreality ("derealization") or being an outside observer of oneself ("depersonalization") or unable to recall important aspects of the event(s) ("psychogenic amnesia"). When those dissociative symptoms occur peritraumatically, the risk of developing PTSD is increased (Bryant et al., 2011). Thus they are the only dissociative symptoms included in the *DSM*-5 version of ASD—with the possible exception of a PTSD intrusive re-experiencing symptom of suddenly feeling as if the traumatic event(s) are happening all over again—flashbacks—which in their most extreme form appear to involve dissociation.

Adaptations for Children in the DSM-5 PTSD Diagnosis

The diagnostic criteria for PTSD in the *DSM*-5 have been modified to include developmentally appropriate indicators for several PTSD symptoms for children ages 7 and above. Notably the re-experiencing symptoms allow for intrusions of traumatically related content without requiring specific details of the traumatic event. Unwanted distressing memories of traumatic event(s) can include repetitive play depicting trauma-related themes, and nightmares can include frightening dreams related in emotional quality or content but without identifiable content. In addition, flashbacks can involve "trauma-specific re-enactment … in play (p. 271)."

The traumatic stressor exposure criterion also may be expanded for children to include the loss of a parent "or other attachment figure" (p. 273). Despite this extended definition, questions remain regarding several adverse experiences of childhood. For example, experiences involving the unexpected or premature death of a primary caregiver fit this definition, but it is not clear whether abandonment by a caregiver—which can have profound adverse effects on child development and adolescent and adult functioning (D'Andrea et al., 2012) and may lead to added trauma exposure or adversities such as multiple out-of-home placements (Ford, Elhai, Ruggiero, & Frueh, 2009) —constitutes a traumatic stressor. Likewise, "betrayal traumas" (Freyd, 1994) that do not involve death, or the threat of death or severe physical injury, such as emotional abuse and childhood neglect, also do not technically constitute traumatic stressors despite clearly constituting serious risks to child health and development (D'Andrea et al., 2012).

A separate PTSD diagnosis for preschool children (ages 6 and under) has been included in the DSM-5. The traumatic stressor (Criterion A), intrusive re-experiencing (Criterion B), and avoidance of reminders (Criterion C) features of PTSD are largely unchanged in the preschool version, except that young children are not expected to have "repeated or extreme exposure to aversive details of [traumatic] event[s]"—that traumatic stressor is primarily expected to occur to adult war combatants or child welfare or emergency response personnel. A major difference is that the negative emotions feature (Criterion D) for preschool children does not include three symptoms from the adult PTSD criteria, each of which is an internal mental state and therefore difficult to detect reliably in children: psychogenic amnesia, negative beliefs about self, and distorted self-blame. The symptom described as "detachment or estrangement from others" in the core PTSD criteria has been modified to focus on behavioral manifestations in the preschool version (i.e., social withdrawal). As a result of the reduced number of Criterion D symptoms and their close relationship to Criterion C avoidance symptoms, preschool PTSD is proposed to require only one symptom from either Criterion C or D. The final symptom feature,

hyperarousal (Criterion E), retains the same five symptoms as in the *DSM-IV* PTSD version, but adds "including temper tantrums" to the anger/irritability item.

Epidemiology

Epidemiological studies, conducted across a variety of populations (Karam et al., 2014; Kessler et al., 2005; McLaughlin et al., 2013) consistently show that exposure to potentially traumatic events (i.e., Criterion A) is quite common, with 60% to 80% of adolescents and adults in international population surveys reporting past exposure to various types of traumatic events, and 6–10% of adolescents and adults having experienced PTSD at some point in their life. Approximately 35% of people who develop PTSD recover within 3 months without treatment (Santiago et al., 2013), but once PTSD has occurred, the individual is at three times greater risk of having a recurrence of PTSD if exposed to another traumatic stressor (Breslau, Peterson, & Schultz, 2008) – thus resulting in many cases of episodically recurrent or chronic PTSD.

Even among young children, exposure to traumatic stressors unfortunately is not a rare phenomenon occurring in the lives of an estimated 25% of children (Briggs-Gowan, Ford, Fraleigh, McCarthy, & Carter, 2010; Costello, Erkanli, Fairbank, & Angold, 2002; Perkonigg, Kessler, Storz, & Wittchen, 2000) to as many as 50–60% of adolescents (Kilpatrick et al., 2003; McLaughlin et al., 2013) in the United States and western European countries. Exposure to potentially traumatic events was even more common among South African and Kenyan 10th graders, affecting more than 80% (Seedat, Nyamai, Njenga, Vythilingum, & Stein, 2004).

In the United States and western European countries, PTSD is relatively rare among young (ages 0–4) children (estimated as about 1 child in every 167) and school-age children (about 1 child in every 100) (Scheeringa, Zeanah, & Cohen, 2010). However, these estimates are comparable to those for current (e.g., in the past 6–12 months) PTSD prevalence in adults (Kessler, Chiu, Demler, & Walters, 2005). Similar current PTSD prevalence estimates have been reported among German adolescents (1–2%; Perkonigg et al., 2000), Puerto Rican children (1%; Canino et al., 2004), and Bangaladeshi children (1%; Mullick & Goodman, 2005). Other studies have reported finding PTSD in as few as approximately 1 in 1000 children and adolescents in Great Britain (Ford, Goodman, & Meltzer, 2003) and Brazil (Fleitlich-Bilyk & Goodman, 2004). Adolescents also are more likely than younger children to meet criteria for PTSD. About 5% of adolescents in the United States (Kilpatrick et al., 2003; McLaughlin et al., 2013) and in Kenya (Seedat & et al., 2004) met criteria for PTSD at some point in their lives.

PTSD is more common in high-risk groups of children, especially those living in impoverished or violent communities. Children living in slums in Bangaladesh were three times more likely than other Bangaladeshi children to have PTSD (3% prevalence; Mullick & Goodman, 2005). Israeli Jewish children, many of whom had been exposed to societal violence but who have not been displaced from their homes, were estimated to have a current PTSD prevalence of 8% (Pat-Horenczyk et al., 2007). In contrast, a much higher prevalence (34%) has been estimated for Palestinian children, many of whom had their homes destroyed and became refugees (Khamis, 2005). Abram et al. (2004) found that most (approximately 90%) of the adolescents in an urban juvenile detention center had been exposed to at least one potentially traumatic stressor (on average, the youths reported experiencing 15 separate incidents during their lives), and 11% met criteria for PTSD. More than 25% of a sample of 10th graders in a country fraught with violence (South Africa) met criteria for PTSD (Seedat et al., 2004).

PTSD is also more common in youth who have experienced multiple traumas. More than half of the teens who report any trauma exposure have experienced at least two different types of potentially traumatic stressors (McLaughlin et al., 2013), and 10–15% have experienced multiple types of traumatic victimization—so-called poly-victims (Finkelhor, Ormrod, Turner, & Hamby, 2005; Ford, Elhai, Connor, & Frueh,

2010). Poly-victims have three times the risk of PTSD, double the risk of depression, and two to five times the risk of substance use disorders and involvement in delinquency or with delinquent peers, compared to other youths—including teens who had experienced traumatic stressors but not as extensively as the poly-victims (Ford, Elhai, et al., 2010).

Comorbidities and Impairment

Epidemiological studies indicate that PTSD is not the only, or even the most likely, psychiatric reaction to follow trauma exposure. Fear, anxiety, sadness, depression, anger, and guilt (among others) are common reactions to traumatic experiences. Other common reactions include physical or somatic complaints, such as health complaints (e.g., gastrointestinal symptoms, headaches), or sleep problems, social and relationship difficulties, and substance use, including alcohol and nicotine (Kessler et al., 2005; Kessler, Sonnega, Bromet, Hughes, & Nelson, 1995; McLaughlin et al., 2013).

Among children (Copeland et al., 2007). PTSD often is accompanied by other anxiety disorders, depression, and behavioral disorders (i.e., comorbidities) and is associated with more severe problems in psychological development, learning and school functioning, peer and family relationships, and, among older children and adolescents, risky and illegal behaviors (D'Andrea et al., 2012; Ford, Elhai, et al., 2010). Even very young children may be significantly affected by exposure to traumatic stressors. Approximately 20% of 18- to 36-month-old toddlers in a US urban sample who were exposed to potentially traumatic events were reported by their parents to have dramatically altered functioning following the event and were described as having higher levels of re-experiencing and arousal symptoms consistent with PTSD (Mongillo, Briggs-Gowan, Ford, & Carter, 2009). In that US urban sample, 2–4-year-old toddlers exposed to interpersonal violence had elevated levels of depression, separation anxiety, posttraumatic stress, and conduct problems, and exposure to potentially traumatic noninterpersonal exposure was associated with phobic anxiety. These associations were not attributable to poverty or parental psychiatric problems, except that parents' anxiety and depression problems mediated the relationship between violence exposure and toddlers' depressive and conduct symptoms (Briggs-Gowan, Carter, et al., 2010).

Etiology and Course

Most children and adolescents who are exposed to traumatic stressors do not develop full PTSD, regardless of age (Briggs-Gowan, Carter, & Ford, 2012; McLaughlin et al., 2013). However, almost half of the children or adolescents who develop PTSD are chronically impaired, i.e., unrecovered as much as three to 4 years after first occurring (Perkonigg et al., 2005). Two trajectories of poor posttraumatic outcomes are most common (Layne et al., 2008): severe initial reactions followed by persistent or episodic posttraumatic impairment (chronic PTSD) and mild to moderate initial reactions followed by severe impairment (delayed PTSD) (Smid, Mooren, van der Mast, Gersons, & Kleber, 2009). Subthreshold or partial syndromes also are more common than full PTSD, and potentially highly impairing, in both the general population and clinical samples of children and adolescents.

Which adolescents, then, are most vulnerable to developing persistent PTSD? The risk or vulnerability factors identified in epidemiological studies of children and adolescents include *pre-traumatic* psychiatric disorder (especially stress/anxiety/affective conditions), impaired caregivers/family, poverty, low IQ, avoidant/anxious coping, generalized arousal and negative affect, female gender, and age (with late adolescents being at highest risk), *exposure* to interpersonal traumatic stressors (e.g., maltreatment, sexual assault, domestic violence) or cumulative (multiple type/polyvictimization or recurrent) traumatic stressors, *peri-traumatic* distress (fear, perceived life threat, dissociation, ASD), and *posttraumatic* low social support, isolation, comorbid psychological problems, poor family

functioning, and cognitive alterations (rumination, distraction, thought suppression, and attention bias toward threat) (Briggs-Gowan et al., 2012; Karam et al., 2014; McLaughlin et al., 2013; Nooner et al., 2012).

Biological factors have been implicated in the vulnerability to developing PTSD, including brain abnormalities such as reduced size or dysregulation of areas in the brain associated with stress reactivity, memory, self-control, and emotion regulation (Kelly et al., 2013; Kuhn & Gallinat, 2013; Teicher & Samson, 2013), alterations in brain-body reactions to stressors (e.g., dysregulation of the hypothalamic-pituitary-adrenal axis as reflected in elevated or depleted cortisol levels), and genetic factors (Teicher & Samson, 2013), as well as learned vulnerabilities that may involve intergenerational (i.e., parent to child) transmission via epigenetic (i.e., changes in genes based on experience) and gene by environment interaction mechanisms. Traumatic victimization, especially in childhood, may result in psychobiological alterations or exacerbation of preexisting vulnerabilities that may sensitize a child or adolescent and thereby increase susceptibility to developing PTSD (Grasso, Ford, & Briggs-Gowan, 2013) and comorbid SUD (Norman et al., 2012).

Traumatic stressors that are deliberately inflicted or interpersonal in nature tend to be more likely than other types of traumatic stressors to result in PTSD and a wide range of associated problems with emotion regulation, interpersonal relationships, and psychobiological development (D'Andrea et al., 2012). Exposure to traumas early in childhood when children are less cognitively developed and more physically dependent on adult caretakers tends to be associated with dissociative symptoms (Dalenberg et al., 2012). Prolonged exposure to traumatic stressors that adversely affect psychobiological development (e.g., childhood abuse, intimate partner violence, genocide and torture) also tend to produce more severe PTSD, SUD, and comorbid emotional and behavioral problems (D'Andrea et al., 2012). Epidemiological findings suggest that youth with multiple types of trauma exposure or extreme emotion dysregulation (e.g., bipolar disorder) also were less likely to *recover* from PTSD once it develops than other trauma-exposed youths (McLaughlin et al., 2013).

PTSD symptoms may not occur simultaneously and will vary over time; one set of symptoms may be manifested with high frequency in one period, followed by other periods that are characterized by a different set of symptoms. Youth often do not make the connection between symptoms (e.g., nightmares, temper flare-ups, depression) and past events or may minimize symptoms in order to convince adults or peers that they are not affected by traumatic experiences. Compared to adult survivors, adolescents may exhibit more impulsive and aggressive behaviors and engage in more traumatic reenactments (i.e., incorporating aspects of traumatic events into their daily lives) (Ford & Blaustein, 2012; Ford, Chapman, Connor, & Cruise, 2012). Behavioral and emotional dyscontrol is more common in adolescents than adults developmentally, consistent with evidence that inhibitory areas of the brain (e.g., prefrontal cortex) mature later than areas related to stress reactivity (e.g., amygdala) and reward seeking (e.g., midbrain).

On the other hand, many children are resilient and either are relatively asymptomatic following exposure to the adverse effects of trauma and able to adapt and preserve both functioning and development (resilient), or have moderate to severe initial reactions or early-onset PTSD (i.e., within 3 months of exposure) but improve and regain normative functioning over time with or without treatment (recovery) (Layne et al., 2008). Resilience in the face of exposure to traumatic or other stressors and a capacity for posttraumatic growth is facilitated by protective factors, such as individual self-control and problem-solving skills, and relational and environmental factors such as good schools, safe neighborhoods, positive relationships with supportive caregivers, non-involvement in risky or undermining relationships, socioeconomic advantage, religious faith, success at school and with peer friendships, older age at time of traumatic exposure, and an absence of prior psychiatric disorders or substance abuse (both by the person and their family) (Copeland, Keeler, Angold,

& Costello, 2010; Milan, Zona, Acker, & Turcios-Cotto, 2013; Wittchen et al., 2012). Individual capacity for resilience in the face of traumatic threat largely depends on human adaptive systems embedding the exposed child in a supportive caregiving web involving caregivers, families, friends, communities, and cultures (Betancourt et al., 2015; Masten et al., 2004). Although the factors associated with preventing versus facilitating recovery from PTSD are not identical (Skeffington, Rees, & Kane, 2013; Sones, Thorp, & Raskind, 2011), they consistently include creating or strengthening of healthy social bonds and resources and psychobiological capacities necessary for adaptive self-regulation (Ford & Blaustein, 2012).

Cultural, Racial, and Gender Differences Contributing to PTSD Symptom Variations

The specific nature of posttraumatic stress symptoms varies for each child, as well as for children living in different ethnocultural contexts. For example, although Kenyan youths were less likely than South African youths to report most PTSD symptoms, they were more likely (50% vs. 30%) to report actively avoiding memories of traumatic experiences (Seedat et al., 2004). Avoidance of reminders of traumatic experiences can be a positive form of coping, but it also can become a part of a vicious cycle in which avoiding memories provides temporary relief but perpetuates a sense of fearfulness that can become full-blown PTSD if minor stressors build up or new traumas occur in the future. Youths who cope by purposefully avoiding reminders of trauma may fare better in the short run, but for some this involves a continuous state of "survival coping" that is a strain on the body, mind, and emotions.

Similarly, there is evidence that racial or ethnic differences influence the development and diagnosis of PTSD (Pole, Gone, & Kulkarni, 2008). However, the impact of these differences on trauma-related disorders or outcomes is unclear because studies rarely report findings by race or ethnicity. Statistics indicate rates of exposure to traumas such as childhood maltreatment and community violence are higher among minorities, especially those living in impoverished communities (Pole et al., 2008). Some studies find that children of ethnocultural minority report higher levels of PTSD symptoms; others do not. In community samples of children (Copeland et al., 2007) and adolescents (Copeland et al., 2007; McLaughlin et al., 2013) in the United States, comparable (and generally slightly lower) levels of PTSD have been reported by Native American, Hispanic/Latino, and African American youth compared to non-Hispanic whites. However, adolescents of Hispanic ethnicity were two to six times more likely than white non-Hispanic youth to report exposure to intimate partner violence and other forms of physical assault; they and African American youth were twice as likely as white youth to report being the victim of stalking and 40% more likely to witness severe injury or death (and for African American youth only, also to experience the unexpected death of a loved one) (McLaughlin et al., 2013). Among adjudicated youths in a juvenile detention facility, Abram et al. (2004) also found no differences in likelihood of PTSD among African-American, Hispanic, and non-Hispanic white youth, and, consistent with the community sample findings noted above, the types of traumatic events to which exposure was reported varied between the ethnocultural groups. Differential rates of exposure to traumatic stressors, lack of access to economic resources, and cultural and societal conditions that either foster violence or undermine protective factors (e.g. social support networks) have been posited as potential factors associated with racial and ethnic differences in children's PTSD (Pole et al., 2008).

Epidemiological studies consistently find that girls are more likely than boys to develop PTSD, as well as internalizing psychiatric disorders that often co-occur with PTSD such as depression (Tolin & Foa, 2006). However, these studies also suggest that this imbalance is not due to girls being more prone than boys to emotionality or affect dysregulation, because female gender was a risk factor independent of the risk conferred by

having a "fear or distress" disorder (McLaughlin et al., 2013). Rather, it may be in part due to differences in the types of traumas that girls tend to experience. Both community and clinical samples show that girls are more likely than boys to experience sexual abuse than boys (Tolin & Foa, 2006). Boys, by contrast, report higher rates of community violence and externalizing behavior disorders such as oppositional-defiant disorder or problems with overt aggression (Herrenkohl, Hemphill, Mason, Toumbourou, & Catalano, 2012; Schwab-Stone, Koposov, Vermeiren, & Ruchkin, 2013)—although one study with youths in mental health treatment for disruptive behavior found that it was girls and not boys who were at risk for externalizing behavior problems if they witnessed either threats (which also was associated with increased risk of internalizing problems only for girls) or actual violence against friends or families (Javdani, Abdul-Adil, Suarez, Nichols, & Farmer, 2014).

Developmental Challenges of Childhood and Adolescent Traumatization and Onset

Exposure to traumatic stressors can lead to fundamental alterations in children's bodies and brains, which are adaptive for "survival coping" (Ford, 2009, p. 33), but which over time can lead children to have serious problems with anger, guilt, shame, grief, dissociation, risky or harmful behavior, damaged or damaging relationships, isolation, and addiction. Perhaps the most harmful effect of persistent posttraumatic survival reactions is failing to develop the abilities necessary for healthy self-regulation. Self-regulation fundamentally involves adaptive engagement in relationships and the ability to utilize and modulate emotions (Ford, 2010).

Although acute stress reactions are normative for children exposed to traumatic stressors, when these symptoms are severe, they also tend to be accompanied by correspondingly severe anxiety and depression symptoms (Barber, Kohl, Kassam-Adams, & Gold, 2014). This constellation of symptoms either in the acute or chronic posttraumatic period is related to problems in developing an adaptive physiological response to cues that signal safety (Jovanovic et al., 2013), thus reducing the individual's ability to reset trauma-related survival reactions and resume adaptive functioning. Studies with adults and children have shown that people with ASD are at risk for PTSD (approximately 33% prevalence) and numerous other psychiatric disorders (approximately 33% additional prevalence) (Bryant, 2011; Bryant et al., 2012). However, most children (and adults) who develop PTSD do not meet criteria for ASD, indicating that ASD is an important early warning sign but by no means an infallible screen for subsequent PTSD.

Although the modifications to the PTSD diagnosis for children included in the DSM-5 (described earlier) may increase the developmental sensitivity of the diagnosis, posttraumatic dysregulation in children does not always take the form of classic PTSD and often involves problems that lead to other comorbid (or alternative) diagnoses—such as reactive attachment disorder (RAD); generalized or phobic anxiety, panic, or obsessive compulsive disorders; bipolar disorder; psychotic or dissociative disorders; eating, body image, or sexual disorders; disruptive behavior disorders; or traits of personality disorders (D'Andrea et al., 2012). While childhood exposure to developmentally adverse traumatic stressors may contribute to the onset of these disorders or exacerbate preexisting symptoms, the most intractable cases (typically children with multiple psychiatric diagnoses) may require treatment addressing traumatic stress reactions in addition to or in lieu of treatment for the psychiatric disorder(s) with which they were diagnosed.

For example, Ford, Connor, and Hawke's (2009) study with psychiatrically impaired children in residential treatment found that, as expected, these children had substantial histories of early life adversity (i.e., documented physical or sexual abuse, impaired parents, multiple out-of-home placements). Cluster analyses revealed distinct subgroups representing different complexities of exposure to these potentially traumatic adversities and showed that the severity of internalizing and externalizing problems could

not be fully accounted for by *DSM-IV* psychiatric diagnoses (nor by age or gender). Two subgroups with complex trauma histories were identified that had severe teacher-rated behavior problems and clinician-rated psychosocial impairment independent of substance abuse, developmental disorders, and externalizing behavior disorder diagnoses. Moreover, substance use disorders were unrelated to behavior problems when the effect of complex trauma was included, and externalizing disorders were associated with impulsivity and hyperactivity only after the effect of a core component of complex trauma—multiple out-of-home placements—was accounted for. Multiple out-of-home placements also contributed to the severity of behavior problems beyond the effects of demographics, psychiatric diagnoses, documented abuse, and parental impairment. This is consistent with the presumed etiology of severe attachment problems, as well as with evidence that children who lose (or never acquire) stable attachment relationships due to abuse or impaired parenting are at risk for long-lasting (at least into early adulthood) problems with affect regulation, disorganized attachment working models, and dissociation (Lyons-Ruth, Dutra, Schuder, & Bianchi, 2006).

Physically abused children also have been observed by adults (Crittenden, 1992) and peers (Salzinger, Feldman, Ng-Mak, Mojica, & Stockhammer, 2001) to be more verbally and physically assaultive than other children. Ford, Connor et al.'s (2009) results suggest that this aggression may not be due to abuse alone, but to survival adaptations based on experiencing the many potentially traumatic conditions that arise in the context of abuse, including parental impairment and out-of-home placements. Further analyses of the data from that sample of psychiatrically impaired children showed that abuse was associated with reactive (not proactive) aggression (Ford, Fraleigh, & Connor, 2010) and with a dampened autonomic response to physical pain (Ford, Fraleigh, Albert, & Connor, 2010). This combination of aggression and reduced physiological responsivity to distress often leads youths to be labeled psychopathic or *callous and unemotional* (CU) and viewed as intractably antisocial. However, girls (Marsee & Frick, 2007) and boys (Kimonis, Frick, Munoz, & Aucoin, 2007) who endorse CU beliefs may be dysregulated consistent with traumatic stress reactions rather than being unempathic and intentionally cruel.

Based on these and other relevant findings (D'Andrea et al., 2012), a complex PTSD diagnosis for children, developmental trauma disorder (DTD), was proposed for (but not accepted in) the *DSM-5*. Results of a survey of more than 1300 pediatric mental health, social work, and medical/nursing professionals internationally demonstrated that symptoms from each of those domains of dysregulation were identified as highly important in assessing and treating children with extensive trauma histories and that developmental trauma symptoms were distinguishable from symptoms of the many other psychiatric disorders with which those children commonly are diagnosed (Ford, Grasso, Greene, et al., 2013; Ford, Grasso, Hawke, et al., 2013). A subsequent interview study with 236 children recruited from mental health and pediatric practices in six US sites (including those with limited or no history of exposure to traumatic stressors or psychopathology as well as those with trauma and psychiatric histories) used a new 15-item DTD semi-structured interview (DTD-SI) which was found to be psychometrically reliable and in confirmatory factor analysis to have a good fit with a proposed 3-criterion structure for DTD with correlated but distinct and internally consistent subscales representing emotional, behavioral and self/relational dysregulation (Ford, Spinazzola, Van der Kolk, & Grasso, 2014, November). DTD's construct validity was supported by hierarchical regressions which showed that, after controlling for *DSM*-IV and *DSM*-5 PTSD, scores for DTD overall and the three factors were associated with parent-rated measures of dysregulation, alexithymia, and impulse control problems. Additional construct validity evidence was provided by logistic regressions showing that PTSD was uniquely associated with exposure to sexual trauma, emotional abuse, and interpersonal violence, but DTD was uniquely associated with family or community violence and an impaired caregiver. Traumatic loss (separation from a

caregiver) was associated with both DTD and PTSD. In terms of comorbidity, the 40% of participating children who met criteria for DSM-IV PTSD (more than half with comorbid DTD) and a comparably large subgroup who met criteria for DTD (also more than half with comorbid PTSD) were two to five times more likely than other participants to meet screening criteria for depressive, manic, psychotic, phobic, separation/generalized anxiety, or obsessive-compulsive disorders. However, DTD, but neither *DSM*-IV or *DSM*-5 PTSD, had three- to fourfold increased risk of *DSM*-IV panic, ADHD, ODD, CD, and eating disorders and *DSM*-5 dysregulation disorders (i.e., non-suicidal self-injury, disruptive social engagement disorder, disruptive mood dysregulation disorder, reactive attachment disorder). DTD thus may comprise forms of dysregulation distinct from PTSD that can be assessed reliably and validly, is associated with violent families and communities and impaired or disrupted caregiver protection, and has more extensive comorbidity than PTSD with other disorders characterized by dysregulation.

Assessment and Treatment of Children's Posttraumatic Stress Symptoms

Assessment of trauma history and posttraumatic symptomatology and functioning most often are done clinically via interview with, or self-report questionnaires or ratings from, youths, parents, other caregivers, teachers, or other knowledgeable informants. For school-age and older children and adolescents, self-report generally can provide a meaningful assessment of trauma history, PTSD, externalizing (e.g., aggression, attention and conduct problems, hyperactivity, substance abuse) and internalizing (anxiety, depression, somatization, dissociation, withdrawal, psychosis, eating and body image) problems and several domains of self-regulation and social competence (e.g., learning problems, daily activities, adaptability, functional communication, leadership, social skills, study skills) (Ford, Nader, & Fletcher, 2013; Frueh, Elhai, Grubaugh, & Ford, 2012; Nader, 2008). For preadolescents and adolescents, it is particularly important to assess risk factors for trauma exposure, exacerbation of symptoms, and serious academic, relational, medical, and legal problems and crises (e.g., suicidality, self-harm, substance use problems, criminal involvement). It is essential to assess age-relevant competences (e.g., impulse control, altruism, consideration, empathy, and responsibility in relation to others, psychological mindedness, executive functions, self-efficacy, and optimism). It is vital to assess these youths' social and physical environment to identify ecological risk (e.g., community or domestic violence, deviant peer group affiliations, modeling of conduct problems by family members) and protective factors (e.g., parental monitoring, responsivity, mutuality, and fostering of autonomy; prosocial peers and activities; adult mentoring). While these risk and protective factors are of particular relevance with older children and adolescents, they should be considered with children of all ages, especially when the child may be in danger of, or actually experiencing, serious physical or psychological harm. In addition to parent/caregiver rating measures, for toddlers and preschoolers, direct observation in home, day care, or clinical settings of parent–child interaction can provide valuable information about the caregiver's and child's strengths and difficulties in self-regulation and relatedness.

Over the past 20 years, several empirically supported psychotherapies have been developed for both acute and chronic pediatric PTSD (Berkowitz, Stover, & Marans, 2011; Chaffin, Funderburk, Bard, Valle, & Gurwitch, 2011; Ford, Steinberg, Hawke, Levine, & Zhang, 2012; Stein et al., 2003). Common treatment elements that are empirically supported include (1) psychoeducation about PTSD, (2) relaxation and coping skills, (3) emotion awareness, expression, and regulation skills, (4) cognitive processing of reactions to trauma, (5) helping the child to construct a therapeutic trauma narrative, (6) in vivo exposure to trauma reminders and practicing of coping skills, (7) conjoint parent-child sessions, and (8) monitoring and enhancing individual safety (Carrion & Kletter, 2012; Ford & Courtois, 2013; Kelly, 2012).

The most extensively researched model of PTSD psychotherapy with children and adolescents is trauma-focused cognitive behavioral therapy (Cohen, Berliner, & Mannarino, 2010; Cohen, Mannarino, & Murray, 2011). TF-CBT includes emotion identification, stress inoculation (e.g., breathing, relaxation) techniques, direct discussion of trauma experiences through gradual exposure exercises, cognitive restructuring, psychoeducation, and safety skill building. Several randomized clinical trials have demonstrated TF-CBT's superiority to supportive therapy with children (including approximately 33% adolescents) with PTSD following abuse, violence, and single-incident (e.g., severe accidents) traumatic stressors (Cohen, Mannarino, & Iyengar, 2011; de Arellano et al., 2014). Outcomes for depression and behavioral problems have been mixed, with moderate effect sizes in some studies (Cohen et al., 2010; de Arellano et al., 2014). When children (or their parents) decline to engage in or cannot seem to emotionally tolerate the processing of specific trauma memories, there are evidence-based therapeutic options for either preparing the child and parents to feel sufficiently safe, confident, and able to modulate distressing emotions so that the child is able to experience the trauma narrative portion of treatment successfully (Cohen, Mannarino, Kliethermes, & Murray, 2012; Matulis, Resick, Rosner, & Steil, 2014), or to enable the child and parent to develop cognitive behavioral (Deblinger, Mannarino et al., 2011) and self-regulation (Ford, Steinberg, et al., 2012; Ford, Steinberg, & Zhang, 2011) skills that address PTSD by reducing everyday traumatic stress reactivity and increasing emotion regulation, self-efficacy, and social support.

Therapies also have been developed to address dysregulation symptoms with traumatized adolescents, particularly when they also are heading for or involved in risky lifestyles and peer groups (e.g., school failure, delinquency, addiction, social isolation, dissociation, self-harm). The most extensively validated self-regulation intervention for youth with PTSD is *Trauma Affect Regulation: Guide for Education and Therapy* (TARGET; Ford & Hawke, 2012; Ford, Steinberg, et al., 2012; Marrow, Knudsen, Olafson, & Bucher, 2012). TARGET teaches a seven-step sequence of self-regulation skills summarized by the acronym FREEDOM. The first two skills, focusing and recognizing triggers, provide a foundation for shifting from stress reactions driven by hypervigilance to proactive emotion regulation. The next four skills provide a dual-processing approach to differentiating stress-related and core value-grounded emotions, thoughts, goals, and behavioral options. The final skill teaches ways to enhance self-esteem and self-efficacy recognizing how being self-regulated makes a contribution to the world. A randomized clinical trial with delinquent or justice-involved girls with dual diagnosis PTSD, substance use and other (e.g., oppositional-defiant, depression, panic) disorders showed that a 10-session individual TARGET intervention was superior to relational psychotherapy in reducing PTSD and depression and improving emotion regulation (Ford, Steinberg, et al., 2012). Additional evidence for TARGET's effectiveness as a group and milieu therapeutic intervention with detained or incarcerated boys and girls was provided by two quasi-experimental studies that showed reductions in violent behavioral incidents and coercive restraints and improvement in PTSD, depression, and hope/engagement in rehabilitation following TARGET (Ford & Hawke, 2012; Marrow et al., 2012). TARGET also has been shown to be effective in enhancing emotion regulation and reducing PTSD with traumatized mothers of young children (Ford et al., 2011).

Five manualized treatments originally developed and extensively validated with adults with PTSD also have been adapted for and empirically evaluated in clinical trial studies with latency-age children and adolescents: prolonged exposure (PE, an exposure-based trauma memory processing intervention; Foa, McLean, Capaldi, & Rosenfield, 2013; Gilboa-Schechtman et al., 2010), cognitive processing therapy (CPT, a cognitive restructuring intervention but includes narrative memory processing; Matulis et al., 2014), eye movement desensitization and reprocessing (EMDR, a modification of exposure and cognitive therapies; Wesselman & Shapiro, 2013), Trauma

and Grief Components Therapy for Adolescents (TGCT-A, a group therapy with exposure and narrative processing components; Saltzman, Pynoos, Layne, Steinberg, & Aisenberg, 2001), and narrative exposure therapy (KIDNET, a modification of exposure and narrative processing therapies; Ruf et al., 2010). In addition, a dyadic mother-child therapy has been validated for young children who have been exposed to violence (child–parent psychotherapy, CPP; Lieberman, Ghosh Ippen, & Van Horn, 2006).

PE utilizes two primary therapeutic tools: imaginal exposure and in vivo exposure. Imaginal exposure involves "revisiting" the most currently distressing traumatic memory, providing a detailed verbal account of the traumatic memory that includes sensory information, thoughts, feelings, and reactions experienced. While recounting their traumatic memory in vivid detail, patients are instructed to verbalize subjective units of distress every 5–7 min on a scale from 0 to 100 or 0 to 10. In vivo activities outside of session involve actually having contact with feared people, places, and things that are reminders of past traumas but that are not currently dangerous. This is done starting with reminders that elicit a moderate level of anxiety (i.e., SUDS of 40) and keeping track of SUDS while remaining in the situation until distress decreases.

CPT involves first writing an impact statement of the personal meaning of the currently most distressing traumatic event, including the effect on beliefs about self, others, and the world, including themes such as trust, safety, self-esteem, and life goals. Subsequent sessions focus on identifying maladaptive thoughts (stuck points), increasing awareness of connections between thoughts and feelings, and processing a traumatic event by writing and reading a detailed narrative account and Socratic questioning designed to help challenge maladaptive thinking patterns while reexamining and elaborating or modifying the trauma memory's narrative description by "weighing the evidence" for and against maladaptive thoughts.

In EMDR, a currently distressing trauma memory is repeatedly described briefly (typically less than 1 min) with titration of the exposure intensity (as in PE) and negative beliefs carefully identified (as in CPT). In addition, a distractor task is used during memory recall (e.g., moving the eyes back and forth or using an alternate bilateral form of audio, visual, tactile, or kinesthetic stimulation). The client then "blanks out" the memory while refocusing on body awareness and breathing deeply. When trauma memory recall is associated with little or no subjective distress, a cognitive exercise called resource installation is conducted, with the client focusing on a positive belief rather than the trauma memory while performing the distractor task. Positive beliefs (e.g., "I can handle this") also are identified and rated on a 7-point Validity of Cognitions scale.

TGCTA is delivered in group sessions that comprise four modules. A first set of sessions address foundational knowledge and skills to enhance posttraumatic emotional, cognitive, and behavioral regulation and interpersonal skills. Next the group processes members' narratives of traumatic events (similar to CPT), followed by additional sessions in which grief and loss narratives are processed in order to facilitate the resolution of traumatic bereavement. A final module has sessions devoted to applying the knowledge and skills to daily life challenges.

KIDNET provides eight individual therapy sessions designed for refugee children with PTSD related to war and other types of organized violence. Rather than focusing only on memories of traumatic stressors, the child is helped to build a chronological narrative of his/her entire life which includes detailed descriptions of traumatic events (similar to PE) and is recorded in writing by the therapist for the child to keep at the close of therapy. Active listening, positive regard, and creative tasks (e.g., constructing a lifeline by placing flowers and stones that represent positive and negative events along a rope to illustrate key events, re-enacting body positions that the child experienced or used to cope during traumatic events) also are used in the narrative work.

Child–parent psychotherapy engages mothers in playful verbal and nonverbal interactions with their infant or toddler child in order to strengthen or restore the mother's capacity to be empathically and responsively available to her child. CPP

therapists do not instruct or correct the mother or child, but instead provide developmental guidance through reflective comments that are hypotheses about what the child may be thinking and intending and the meaning that this has for the mother in light of the tension between her own posttraumatic reactions and her caring and affection for her child. CPP therapists also model appropriate protective behavior, provide the mother with emotional support, and assist her with crisis intervention and practical problem solving when stressors occur in the family's current life. CPP therapists pay special attention to helping the parent and child understand the impact of traumatic events on their individual and shared experience, as well as to access and share memories of positive experiences together (and for the mother, experiences of being cared for as a child or adult by her own parents) in order to sustain the mother as she cares for her child and both recover from the impact of trauma.

Several other promising psychotherapies for adolescents with complex PTSD/DTD have been developed (Ford & Courtois, 2013) or adapted from adult versions (Courtois & Ford, 2009), including *Trauma Recovery and Empowerment Model* (Ford, Fallot, & Harris, 2009), the *Sanctuary Model* (Bloom, 2013), *Trauma Systems Therapy* (Navalta, Brown, Nisewaner, Ellis, & Saxe, 2013), *Dialectical Behavior Therapy* (DBT; DeRosa & Rathus, 2013), *Structured Psychotherapy for Adolescents Responding to Chronic Stress* (SPARCS; Ford, Blaustein, Habib, & Kagan, 2013), *Attachment/ Regulation/ Competence* (*ARC*; Ford, Blaustein et al. 2013), and Skills Training in Affect and Interpersonal Regulation (Gudino et al., 2014). *Seeking Safety* (Najavits, Gallop, & Weiss, 2006; Najavits & Hien, 2013) is a psychotherapy for adolescents that addresses PTSD and comorbid substance use disorders. Two school-based group educational and therapeutic interventions have shown evidence of effectiveness with inner city children (Cognitive Behavioral Intervention for Trauma in the Schools, C-BITS; Stein et al., 2003) and with middle school children experiencing the ongoing threat of war in the Middle East (ERASE-Stress; Berger, Gelkopf, & Heineberg, 2012).

Pharmacological interventions are often considered as adjunct to psychotherapy for PTSD. Medications can reduce symptoms when impairment disrupts daily functioning or helps the child tolerate emotional pain associated with traumatic memories that may be augmented by psychotherapy (Connor, Ford, Arnsten, & Greene, 2014). Selective serotonin reuptake inhibitors (SSRIs) are often chosen for treating pediatric PTSD, but Cohen Mannarino, Perel, and Staron (2007) found no evidence in a randomized clinical trial of added benefit when a SSRI (sertraline) was combined with TF-CBT for 10–17-year-old youth who had histories of sexual abuse. A 10-week randomized controlled trial using an independent sample sertraline (50–150 mg/day, mean dose 115 mg/day) did not offer any advantage over placebo on a measure of PTSD symptom severity (Robb, Cueva, & Sporn, 2010). Although SSRIs may have a role in treating anxiety and depressive disorders comorbid with PTSD, their efficacy for children with PTSD is not evident. Adrenergic medications such as guanfacine extended release (which has shown promise in an open trial for traumatized children's daytime hyperarousal) and prazosin (which has demonstrated efficacy in reducing adults' PTSD hyperarousal) warrant clinical trial studies (Connor et al., 2014). No systematic studies of antipsychotics or mood stabilizers have been reported with children with PTSD (Connor et al., 2014). For children with ASD due to burn traumas, tricyclic and SSRI antidepressants have shown inconsistent evidence of efficacy, but a chart review study found a correlation between average morphine dose and amount of decrease in PTSD symptoms. Thus, pharmacologic pain control may help mitigate against ASD when traumatic events involve physical as well as psychological injury (Connor et al., 2014).

Early Identification of Traumatized Children and Prevention of PTSD

A recent national survey of adults found that the median age of PTSD onset was 23 years of age (Kessler et al., 2005). Thus, trauma-exposed children and adolescents who do not have PTSD will

develop PTSD in adulthood unless preventive interventions are provided to those at risk. This further underscores the importance of identifying and delivering evidence-based treatment to adolescents with PTSD before they reach adulthood, as well as prevention interventions for children and adolescents who are at risk for PTSD on a selective basis before posttraumatic symptoms emerge or on a targeted basis before posttraumatic symptoms crystallize into a disorder involving serious developmental and psychosocial impairment.

Although the development of interventions designed to prevent PTSD following trauma exposure is an emerging area, several approaches have been examined in the adult and child literature. Most selective and targeted prevention interventions include one or more of the following therapeutic approaches: (1) psychoeducation to normalize posttraumatic stress reactions and symptoms, (2) debriefing to promote emotional processing of the experience and impact of exposure to traumatic stressors, (3) psychological first aid, (4) increased communication within the family, and (5) enhanced coping skills (Marsac, Donlon, & Berkowitz, 2014). However, the evidence in support of each of these individual components is inconsistent.

Psychoeducation, for example, is a common component of many preventive interventions, but it has not been sufficiently studied independently to determine its comparative effectiveness in preventing PTSD (Forneris et al., 2013; Pfefferbaum, Newman, & Nelson, 2014). Psychological debriefing in the immediate aftermath of a traumatic event had been widely espoused as a method of reducing distress and preventing PTSD. However, several critical reviews of randomized controlled studies of adults have concluded that this approach does not prevent PTSD and may, in fact, lead to an increased rate of PTSD by interfering with the natural recovery process (Forneris et al., 2013; Kearns, Ressler, Zatzick, & Rothbaum, 2012). A controlled study of the use of psychological debriefing with children following a motor vehicle accident likewise found no benefit (Stallard et al., 2006). A review of debriefing studies with children found them to be methodologically inconsistent in terms of what constitutes "debriefing" and the timing of the intervention, with some studies describing delayed delivery up to several months following the traumatic event. Outcomes were also inconsistent, with some studies finding positive effect, and others no effect, although none found it to be harmful (as some adult studies have) (Pfefferbaum, Newman, et al., 2014). Although psychological first aid appears to be a promising intervention delivered in the immediate aftermath of disasters, it has not been well studied, and therefore its efficacy has not been empirically documented (Marsac et al., 2014). However, its components (promoting safety and comfort, stabilization, connecting individuals with social and community supports, providing practical assistance, offering information about responses to trauma and coping) are each evidence informed by current knowledge on the prevention and treatment of trauma.

Among adults, the administration of cognitive behavioral therapy (CBT) within the first few weeks following trauma exposure to individuals displaying symptoms of acute stress disorder has been shown to be effective at preventing the onset of PTSD compared to no treatment or supportive counseling (Forneris et al., 2013; Kearns et al., 2012). Although similar comparative studies of the early provision of CBT have not been conducted with children, two studies have evaluated stepped preventive care interventions that adjust the level of intervention according to each individual's needs, which included components of CBT and other approaches to promoting coping resources.

A stepped collaborative care model that has been found to be effective with adults (Zatzick et al., 2004) was adapted by Kassam-Adams et al. (2011) as a selected prevention intervention with hospitalized injured children. Following universal screening, children identified at risk of developing PTSD were randomized to receive usual care or an intervention consisting of psychoeducation and assessments to identify needs and, as needed, one or more of the following elements: assistance with care coordination, medical adherence, and communication with medical

providers, a brief parent-child intervention, further evaluation by a mental health provider, and TF-CBT. However, no differences in PTSD or depression symptoms emerged between the intervention and usual care groups. Zatzick et al. (2014) provided stepped collaborative care to injured adolescents during the year after screening and assessment for PTSD in the emergency room. Severe PTSD, depression, or alcohol and substance abuse symptoms were rare in this sample and were not improved more by the collaborative care than by usual care. However, fully one third of these teens carried weapons at the time of their injury, and by the one-year follow-up assessment, only 7% receiving collaborative care were still carrying weapons (compared to three times as many, 21%, of youth receiving usual care. This reduction in gun carrying also was observed with youth who had sustained a traumatic brain injury.

Child and Family Traumatic Stress Intervention (CFTSI; Berkowitz et al., 2011) is a four-session child and caregiver intervention conducted within 30 days of a potentially traumatic event that utilizes psychoeducation and behavioral and cognitive approaches to managing traumatic symptoms with the aim of increasing familial support, parent-child communication, and coping resources. A randomized controlled trial comparing CFTSI to a four-session supportive intervention found that intervention group had lower rates of PTSD and fewer traumatic stress and anxiety symptoms at 3-month follow-up (Berkowitz et al., 2011).

Thus, although the extensive intervention provided by stepped collaborative care may not be necessary if injured children are provided with effective medical care, a briefer focused PTSD prevention intervention for the child and family shows promise with acutely traumatized children and youth. It also is possible that stepped collaborative care may be needed and effective, if the subgroup of acutely injured children and youth who are experiencing severe PTSD, depression, or alcohol/ substance use problems are targeted rather than evaluating the intervention on a selective basis. The striking finding of reduced carrying of weapons by teens in collaborative care suggests that this intervention may have a positive impact with highly hypervigilant youth.

The ability of pharmacological interventions to prevent the onset of PTSD in youth who are hospitalized following physical injuries has also been examined. Studies of morphine use with injured children and adults suggest that pain reduction may contribute to preventing PTSD (Marsac et al., 2014). Sheridan et al. (2014) re-assessed children and adolescents 4 years after hospitalization for severe burns and found that PTSD symptoms were inversely correlated with the dose of morphine provided during acute burn care. While promising, these findings do not demonstrate that opiate (morphine) treatment effectively prevents PTSD, but instead indicate that medical care for children who are in physical pain should carefully monitor and take timely steps to reduce the pain to tolerable levels in order to reduce the long-term severity of PTSD symptoms. The risk of chronic addictive opiate use after acute use of opiates in hospital care also requires extreme caution (Holman, Stoddard, & Higgins, 2013).

A randomized controlled trial of sertraline use with severely burned children documented a decrease in children's PTSD symptoms as compared to placebo by parent report, but not by child report of symptoms (Stoddard et al., 2011). Although studies of the use of propranolol in adults have had mixed results, with some finding that administration soon after trauma exposure reduced later PTSD severity, studies with children have not demonstrated its efficacy (Kearns et al., 2012; Nugent et al., 2010; Sharp, Thomas, Rosenberg, Rosenberg, & Meyer III, 2010).

In addition to the selective and targeted early intervention approaches described above, resilience-enhancing pre-trauma interventions aimed at preventing PTSD are also emerging. A recent review identified seven published studies of these primary prevention programs for adults (Skeffington et al., 2013). All programs included psychoeducation on the nature and impact of exposure to life stressors and expectable stress responses, and a subset also included skill-building components such as relaxation techniques, cognitive coping, attentional control, and emotion regulation. However, methodological shortcomings precluded

the authors from drawing any conclusions as to the efficacy of pre-trauma exposure primary prevention programs for PTSD. Among children, Pfefferbaum, Varma, Nitiema, and Newman (2014) identified five studies of pre-disaster preparedness interventions for children (three from the same group of investigators) largely conducted in school settings using a combination of psychoeducation and cognitive-behavioral techniques to increase disaster awareness, prevent hazards, and/or build resilience, which resulted in improved posttraumatic stress outcomes.

Conclusion

The DSM-5 has increased the developmental and diagnostic sensitivity of the PTSD criteria by expanding the stressor criterion, including developmentally appropriate indicators, and refining the scope and definition of symptoms to better reflect trauma-related impairment in self-regulation. Better understanding of comorbidities, risk and protective factors, comorbidities, and the amplified impact of experiencing exposure to multiple types of traumatic stressors has further refined the conceptualization, assessment, and treatment of pediatric PTSD. Several empirically supported psychotherapies for pediatric PTSD have been developed and disseminated in the past 20 years. Finally, as approaches to early identification and intervention with traumatized children develop and accrue evidence, the prevention of PTSD and ASD is an emerging possibility.

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Part VIII

Internalizing Disorders: Feeding and Eating Disorders

Pica

16

Andrea S. Hartmann and Verena Jurilj

Case

Alexia is a 13-year-old girl. She is physically healthy and good at school and gets along well with her parents. She reports having friends and is hanging out with them frequently, whereas her mother describes concerns about Alexia being a loner socially. Alexia's mother brought her in for a consultation because over the past 2 months, Alexia has often locked herself in her room for periods of time and overall has seemed withdrawn and depressed. Upon direct conversation with Alexia, she admits retreating into her room but denies feeling lonely or dysphoric. She describes that she retreats into her room when she feels the urge to eat eye shadow. She seems embarrassed to explain the process, but after some prompting, she reports buying several compacts of eye shadow daily in the local drug store. She has a preference for a specific brand, in particular for the taste of the brands' gray color. While putting on eye makeup about 3 months ago, some makeup dropped onto Alexia's lips. She liked the taste and consistency so much that she continued eating it. Because her worried mother now watches her more closely, Alexia often tries to withstand the strong urge to eat eye shadow, but always gives in to her cravings after a few hours. Although Alexia has not told anyone about her makeup consumption, she assumes that no one she knows shares the habit. Having disclosed her information during the consultation, she reports feeling ashamed. She believes that people would think she were crazy. On the other side, she worries that the eye shadow could harm her health.

Symptom Presentation

Alexia is a typical example of an adolescent with the feeding and eating disorder pica. Other than in individuals with quantitative feeding and eating disorders such as anorexia nervosa, bulimia nervosa, and binge eating disorder that are characterized by eating too much or too little, individuals suffering from the qualitative feeding and eating disorder pica regularly crave and intentionally consume nonnutritive, nonfood items (American Psychological Association [APA], 2013). They usually do not have any aversion to food in general. The range of possible items consumed is vast, including earth, clay, hair, paint, cigarette butts, laundry starch, paper, chalk, and feces. As Young (2011) points out, the substances are often dry or even powdery. When reading

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online first-person accounts (e.g., Humhoney, 2010), individuals are usually very specific about cravings (e.g., consistency and the composition of the item, where it is acquired, or how it smells and feels). They might favor a specific type of laundry starch that has just the right consistency, or crave a specific kind of clay after it has been baked for the right duration to adjust its dampness. Very little is known about the amount of substances usually consumed, probably due to difference in individual and substance. For earth eating (called *geophagy*), the typical amount consumed seems to be 25–45 g daily, depending on the substance's consistency and the population (e.g., schoolchildren, Geissler, Mwaniki, Thiong'o, & Friis, 1997; pregnant women, Luoba et al., 2005). Individuals with pica experience strong cravings toward the substance they consume. Just like Alexia, they usually feel a strong urge to consume the substance, and even when they try to withstand it, they usually give in.

For children suffering from pica, it might be even harder than for adults to control their urges as inhibition mechanisms might not yet be fully developed. There is no data comparing the prevalence of consumption of different pica substances, particularly for children and adolescents. Most of the prevalence data for children was collected in areas where geophagy was the most common pica substance (e.g., Nchito, Geissler, Mubila, Friis, & Olsen, 2004). Other studies found plaster or paint chips (De la Burdé & Reames, 1973; Marchi & Cohen, 1990) to be prevalent. In addition to those findings, other substances listed in previous versions of the Diagnostic and Statistical Manual of Mental Disorder that are particularly typical for infants include string, hair, and cloth, while outdoor substances comprise animal feces, stones, and insects (APA, 1987).

Looking at the abovementioned online first-person account and others, it seems that many individuals suffering from pica know about the medical consequences their disorder consumption can create. Some individuals notice problems with their digestion or skin, but cannot stop eating their desired items. Up to a certain age, however, children may not be cognitively able link possible consequences to their pica or might not even notice them. Thus, treatment of pica in children could pose a great challenge if they are not able to recognize the negative implications their behavior has.

Individuals who struggle with pica often hide their cravings and consumption from their family and friends because they feel embarrassed. Young (2011) reported after analyzing posts found in an online discussion group about cornstarch pica: individuals may secretly order large amounts of their craved item on the Internet (e.g., pencils, starch, baby powder) or sneak outside to taste the earth from the nearby forest after the rain. Everyday items like toilet paper or tissues might also be consumed, only raising the suspicion of the family members when consumed in huge amounts. Sometimes, the location of the acquired item is chosen to conceal their behavior, such as stripes of wallpaper behind a cupboard might be ripped off so that the missing wallpaper is less likely to be noticed than in other areas. Particularly children and adolescents may hide it for fear of sanctions or closer surveillance.

Like Alexia, many individuals are unlikely to report their pica behaviors without specific inquiry, as they do not consider it worth reporting or feel embarrassed (Rose, Porcerelli, & Neale, 2000). In a recent study, in- and outpatient patients being treated for eating disorders or weight loss were specifically asked about pica eating behaviors. Of those reporting such eating habits, some stated they were embarrassed, and that until this interview, no one had inquired about such behavior (Delaney et al., 2014).

History of the Diagnosis of Pica in the Diagnostic and Statistical Manual of Mental Disorders

Before inclusion in the Diagnostic and Statistical Manual of Mental Disorders (DSM), in the medical context, pica first appeared in 1563: Geophagy was described in a medical book in pregnant women and in children (Rose et al., 2000). Prior to the twentieth century, pica was usually not seen as an independent disorder (Hakim-Larson, Voelker, Thomas, & Reinstein, 1997), but had

commonly been subsumed under other diagnoses such as anorexia nervosa, bulimia, and rumination (for a more extensive historical review of the diagnosis, see Parry-Jones & Parry-Jones, 1994).

Pica first appeared in the DSM with the release of the third edition of the DSM (DSM-III) by the APA (1980). Found in the "Infancy, Childhood and Adolescence Disorders" section at that time, the diagnostic criteria for pica included the repeated consumption of a nonnutritive substance that was not due to another mental or a physical disorder (e.g., infantile autism or Kleine-Levin syndrome). The eating habit had to exist over a period of at least 1 month. The usual age of onset reported was 12–24 months, with the disorder rarely persisting into adolescence or adulthood. Some predisposing factors were listed, including mental retardation, mineral deficiencies, and family-related issues (e.g., neglect or poor supervision). The listed complications associated with the disorder were lead poisoning and intestinal obstruction.

In the revision of the DSM-III (DSM-III-R; APA, 1987), the diagnostic criteria remained the same, as did the category, then called "disorders usually first evident in infancy, childhood, or adolescence." Mineral deficiencies and family-related issues were not listed as predisposing factors. The DSM-III-R categorized three populations with pica cases that today still represent the groups of individuals in which pica is most prevalent (see "Epidemiology and Course"): young infants, individuals with mental retardation, and pregnant women.

The fourth edition of the DSM (DSM-IV; APA, 1994) added some complexity to the diagnostic criteria of pica by requiring that the eating habit being inappropriate to the individual's developmental level and not a part of a culturally supported practice. The latter has been reported frequently, including in a report by Abraham and Parsons (1996) that found women in tropical latitudes engaged in geophagy around their pregnancy because they believed the soil to increase fertility. Sensitivity to culture practice has been criticized repeatedly. For example, Paniagua (2000) posits that such an emphasis on cultural variables is uncommon for the DSM and argues that the exclusion of a great number of individuals with pica behavior might result in a failure to detect individuals with severe and life-threatening conditions. For the first time, the DSM allowed for comorbid diagnosis of pica with other mental disorders if the pica was severe enough to warrant clinical attention. The DSM-IV also listed typically consumed items, varying with the age of the individual concerned. Compared to the earlier versions of the DSM, a more detailed description of the course and complications of the disorder had been added, but the limited knowledge about the disorder has also been emphasized. Also, pica was included as a disorder seen in adults for the first time, having been moved from the "Feeding and Eating Disorder of Infancy or Early Childhood" section to the "Eating Disorder" section as an example of "Other Eating Disorders." In the text revision of DSM-IV (DSM-IV-TR; APA, 2000), the diagnostic criteria did not change, and only descriptions of associated disorders and prevalence data for a subgroup of adults with mental retardation were added.

Current DSM-5 Diagnostic Criteria

In the DSM-5 (APA, 2013), pica moved as a full syndrome, not only as an example of "Other Eating Disorders," to a new combined category "Feeding and Eating Disorders." Also, some specifications were made to clarify the diagnostic criteria. First, the substances consumed in pica are now specified as "nonfood" in criterion A. Therefore, nonnutritive substances regarded as foods or beverages are no longer items warranting a pica diagnosis. Examples of such substances which raised discussion in the past are "diet" beverages that have no calories and therefore no nutritional value. Also, ice intended for eating (e.g., ice cubes or crushed ice) no longer warrant a diagnosis of pica even if consumed in unusually large amounts. Cornstarch, uncooked rice, and other food items usually in need of processing prior to consumption are now excluded from the diagnosis as well, but were considered pica substances before. However, ice and starch not meant for eating, e.g., frost scraped from the

interior of a freezer and laundry starch, still count as pica items.

Although criterion B has not changed, there are new specifications in the "diagnostic features," describing that pica should only be diagnosed in individuals at least 2 years of age. Any behavior similar to pica before 2 years of age may represent developmentally appropriate mouthing of objects, sometimes leading to inadvertent ingestion. Specifying this age limit is in line with the criteria in the International Classification of Diseases 10th version (ICD-10; World Health Organization, 1992).

Criterion C added the provision that in order to confer a diagnosis of pica, the eating behavior should not be part of a socially normative practice (see description of culturally supported practice in DSM-IV). In addition, criterion D specifies that the pica diagnosis should be given in the context of another mental disorder or medical condition only if needing additional clinical attention. The term medical condition also includes pregnancy. The craving of unusual food items can be common in pregnancy, to the extent that in some regions, a woman may be considered pregnant if she starts to show pica behavior (Hunter, 1993). Thus, the necessity for additional clinical attention needed before adding a pica diagnosis might prevent stigmatization and pathologization of whole subpopulations.

Examining Alexia's case from the beginning of the chapter, she meets the DSM-5 diagnostic criteria for pica: She describes frequent consumption of eye shadow, clearly a nonnutritive, nonfood substance, over a period of more than 1 month (A). She is an otherwise mentally and physically healthy adolescent (D), so the eating of eye shadow is inappropriate to her developmental level (B). She lives in an urban, US family, where the consumption of makeup or any of the main components of her eye shadow is neither a culturally nor a socially normative practice (C).

Epidemiology and Course

To our knowledge, there are no community-based prevalence studies of DSM-5 pica. There are only a few studies that report prevalence of pica among representative populations of children, dating back to 1942 (Dickins & Ford, 1942) and most recently 2004 (Nchito et al., 2004). Of course, comparability and generalizability of these data is limited due to different diagnostic criteria used. However, throughout the literature, children are one of the three major populations presenting with pica, along with pregnant women and individuals with intellectual or developmental disabilities. Rose et al. (2000) summarized that pica seems to be most common in underdeveloped areas (e.g., rural African populations; Young, 2011) with low socioeconomic status, in women (especially pregnant women), and in children, with prevalence rates generally decreasing with age.

Epidemiological studies in youth were mainly conducted in the USA and in different regions of Africa, yielding prevalence rates ranging from 1.7% for children in upstate New York counties (Marchi & Cohen, 1990) to a considerable 74.4% in children in Zambia (Nchito et al., 2004). However, these studies have to be interpreted with caution. Several studies conducted in the USA employed criteria different from those of the DSM (e.g., consumption of two or more nonfood substances (Marchi & Cohen, 1990)), specific subgroups (e.g., children being treated for sickle cell disease (Ivascu et al., 2001), children currently treated in clinics (Millican, Layman, Lourie, Takahashi, & Dublin, 1962)), or age ranges including toddlers whose behavior was still developmentally appropriate and thus would not meet DSM criteria for pica (e.g., De la Burde & Reames, 1973; Marchi & Cohen, 1990). Young (2011) summarized in her review of the literature that highest prevalence rates for children are found in African countries and in the US regions with low socioeconomic status. The studies in African populations show that children mostly engage in geophagy, while studies conducted in other countries (mainly in the USA) report different forms of pica (e.g., paint, paper). There is evidence, however, that pica represents a socially appropriate behavior in certain African regions, which would result in dismissal of a pica diagnosis in many cases if diagnosed according to DSM-5 criteria. For example, Geissler et al. (1997) report that in their study of Western Kenya, "soil eating is a conscious and

non-stigmatized activity of the children in the study area and part of their collective everyday life" (p. 625). Another study reported that in Zambia, local vendors sold earth for consumption (Nchito et al., 2004). In sum, many prior studies have used criteria disparate from the DSM and reported on specific subgroups that are not translatable to our current clinical definition of pica. Further research using rigorous methodology consistent with the current DSM (DSM-5) is needed.

In the pregnant women and individuals with developmental or intellectual disabilities, Young (2011) reports 47 representative studies (*n*'s ranging from 40 to 70,000). The most recent of the studies in pregnant women (utilizing DSM-IV-TR criteria) reported prevalence rates from 0.02% in a national representative sample in Denmark (Mikkelsen, Andersen, & Olsen, 2006) to as high as 63.7% in a health facility sample in Tanzania (Nyaruhucha, 2009). Geophagy was again the most prevalent form of pica in African samples. Among individuals with intellectual disabilities, Ashworth, Hirdes, and Martin (2009) reported that 22% or more of 1000 institutionalized individuals with intellectual disabilities in the USA suffered from pica, using DSM-IV-TR criteria. Zainab (2001) reported high prevalence rates in institutionalized populations (9–25%) and slightly lower rates in community settings (0.3–14.4%) in his review. The large range of these prevalence rates might be due to a sampling bias or be due to an increase with mental impairment.

In sum, as recent epidemiological studies are still missing, no clear conclusion can be drawn for the prevalence rates of pica among children (other subgroups or the general population). The prevalence rates of all available studies require cautious interpretation, as some studies date back to the mid-twentieth century, using outdated, less specific diagnostic criteria than the DSM-5 criteria at the time or describing pica behavior, not the disorder (see above). For instance, in all subgroups, the new categorization as nonfood (particularly for ice cube and raw starch pica) might diminish the frequency of pica cases.

Although there is still a lack of longitudinal, representative data, the typical onset of the disorder seems to be in childhood, often spontaneously remitting after a while and rarely persisting through adolescence and adulthood (APA, 2013). This is reflected in the prevalence rates that seem to decline with age except for certain subpopulations, as mentioned above. However, in children with developmental or intellectual disabilities, pica can persist over time if it remains untreated, as difficulties in perception and learning as well as resulting challenging behaviors do not remit in most people with such disabilities (Matson, Belva, Hattier, & Matson, 2011). Moreover, pica may onset in adolescence or adulthood, particularly for certain subgroups like pregnant women (APA, 2013).

Comorbidities, Associated Impairments, and Developmental Challenges

As previously mentioned, pica (or pica behavior) can be associated with intellectual or developmental disorders, e.g., autism spectrum disorder (ASD). A recent study reported pica behavior in half of its sample of the children (3–10 years of age) with ASD (Clark, Vandermeer, Simonetti, & Buka, 2010), supporting the findings of an earlier study (Kinnell, 1985) that found that 60% of the individuals of the ASD group had shown pica behavior once and 13% repeatedly to chronically. Pica behavior in ASD might be seen as part of a prolonged developmental delay (oral investigation and mouthing behavior are common in healthy toddlers, as mentioned above) and therefore might not warrant a separate diagnosis. However, one should consider the need of additional medical attention due to potential medical consequences, potentially rendering a diagnosis useful.

In adults, other comorbidities can be found as well. Bhatia and Gupta (2009) reported a case where a woman described an impulse to eat chalk. Her pica eating was similar to obsessions (egodystonic and persistent) and decreased with education about coping mechanisms during stressful situations and the use of a selective serotonin reuptake inhibitors (SSRIs). Thus, pica may in some cases be stress-induced and might share underlying mechanisms with obsessive-compulsive spectrum disorders (Bhatia & Gupta, 2009). This supports

older findings by Stein, Bouwer, and van Heerden (1996) that analyzed five cases of pica. Two cases described pica behavior as a compulsion, while two other cases describe pica behavior as a means to decrease tension, similar to an impulse control disorder. Four of the cases were successfully treated with SSRIs. Dumaguing, Singh, Sethi, and Devanand (2003) reported on three cases of geriatric patients with schizophrenia or schizoaffective disorders who developed pica late in life, resulting in fatalities in two of the cases (asphyxiation and obstruction/punctuation). In a field study for DSM-5 criteria of feeding and eating disorders, Delaney et al. (2014) found prevalence rates of pica behavior of 7.4% in a residential eating disorder sample and 4.0% in an outpatient weight-loss seeking sample with most patients reporting pica behaviors for the reason of weight control. Only 1.3% (two individuals) of the residential sample (and 0% of the weight-loss sample), however, received a DSM-5 pica diagnosis consuming gum and plastic.

Besides comorbidities with other psychological disorders, pica is associated with a number of medical issues. Decker (1993) reports that of 35 patients who received treatment for pica in a hospital, three quarters required surgery, with one third resulting in complications and 11% resulting in death. As children might not be able to link pain and digestive problems to their pica behavior, they might develop even greater medical impairments before pica is noticed and treated. As our digestive system has adapted to certain types of food, it cannot cope with most nonfood items, and depending on the substance, there may be various resulting medical complications. Such complications include intestinal perforation due to the shape and consistency of the objects ingested (e.g., rocks, metal parts), which might result in the need for surgery, or even death. Other possible complications include intestinal obstructions (e.g., when hair is consumed) or lead poisoning (e.g., when paint is consumed). Furthermore, endoscopies and laparotomies might be necessary to remove nondigestible foreign objects from the intestinal tract (Decker, 1993). Asphyxiation is also possible when swallowed items clog the trachea (Dumaguing et al., 2003). Infections are also frequent when earth is the preferred substance: For example, Luoba et al. (2005) examined the reinfection rate with intestinal helminths among women in Kenya who were dewormed during their pregnancy and found that women with geophagy were more often and more intensely reinfected postpartum than women without geophagy. Saathoff, Olsen, Kvalsvig, and Geissler. (2002) reported similar results among schoolchildren in South Africa who ate soil from termite mounds. Many individuals with pica also have deficiencies in minerals or vitamins, although this is often considered a preexisting or even risk factor rather than a consequence of the disorder (see "Etiology"). There is not much literature on the psychological consequences of pica, but many individuals have reported feeling significant levels of shame (see "Symptom Presentation"). As a consequence, individuals struggling with pica may socially withdraw and/or develop depressive symptoms.

Pica may be associated with developmental challenges, but research has not targeted this topic to date. First, one might assume that the association with intellectual disability and other neurodevelopmental disorders might delay the individuals concerned in all aspects of development. This factor might be particularly pronounced given the large comorbidity between pica and developmental or intellectual disabilities (Matson, Hattier, Belva, & Matson, 2013). Second, social withdrawal due to shame may lead to depression, which is often associated with developmental challenges (Steinhausen, 2013). Finally, pica may lead to medical consequences and residential/emergency treatment including long hospital stays, which can impact children and adolescents developmentally, e.g., in their school or social development.

Differential Diagnoses

There are five main differential diagnoses one has to consider when diagnosing pica. If the pica behavior only occurs in the context of another disorder or serves a purpose within another condition, the diagnosis of pica is not warranted.

First, if nonfood, nonnutritive items are solely consumed in order to lose weight or prevent weight gain (e.g., in eating disorders), then a diagnosis of pica is not warranted. In a sample of 149 patients diagnosed with eating disorders in a residential treatment center and 100 weight-loss seeking patients in an outpatient clinic, Delaney et al. (2014) found that while only two patients met the DSM-5 criteria for pica, a total of 15 patients engaged in pica behavior. Qualitative analysis showed nine of those individuals mainly ate ice, for different reasons (e.g., as a supplement for food, to keep the mouth moist when restricting fluids, or to avoid hunger). Second, individuals suffering from avoidant/restrictive food intake disorder may concurrently exhibit pica behavior, but a separate diagnosis of pica would not be conferred, as they may actively seek out substances with the preferred sensory (i.e., textural, smell) qualities irrespective of them being considered food or having nutritional value (Hartmann, Becker, Hampton, & Bryant-Waugh, 2012). Third, individuals who engage in nonsuicidal self-injury and ingest nonfood items might swallow potentially harmful objects, like razorblades, or objects that cause pain during digestion, like stones, to evoke pain as an emotion regulation strategy. Ingestion of such objects is thus treated as part of the disorder (i.e., treatment of self-injurious behavior) and does not warrant an additional pica diagnosis. Fourth, some individuals with schizophrenia (e.g., Fishbain & Rotondo, 1983; Foulon, 2003) may consume nonfood substances, possibly as a consequence of auditory hallucinations in which the voices order the individual to do so or as a function of a delusional system. In these cases, pica behavior may cease once overall schizophrenia symptoms are treated. Finally, DSM-5 mentions factitious disorder is named as a possible differential diagnosis. In factitious disorder, individuals swallow and ingest items to cause injury and falsify medical symptoms in order to get medical attention. Pica behavior is part of the deceptive behavior typical of the disorder and therefore not diagnosed separately (APA, 2013).

In addition to the aforementioned presentations that may include pica behavior, there are other presentations that may consist of similar characteristics. For instance, pica may be falsely diagnosed as a substance use or addictive disorder; from a clinical perspective, the cravings of individuals with pica are comparable to that of individuals struggling with substance use. Internet forums for people suffering from pica (e.g., http://www.experienceproject.com/, search for pica) contain many examples of language very similar to that of individuals abusing drugs: "its dry, white and powder texture drives me insane" (Adiaz268, 2011), "I (…) somewhat panic when I run out" (Omma41, 2014), "I didn't give in for a long time. One day (…) I couldn't take it anymore. I went outside and got a rock (clay) and put it in my mouth. It was so satisfying" (Humhoney, 2010), "I've been clean", and "I did…" (Young, 2011, p. 14f). Some criteria for addiction might also be fulfilled by someone suffering from pica (e.g., the time spent to obtain the specific craved substance), but tolerance effects are likely not seen with pica. Therefore, if the substance consumed is psychotropic and listed in the "Substance-Related and Addictive Disorders" category of DSM-5, pica should not be diagnosed. One might also see a resemblance to the attempts by individuals suffering from obsessive-compulsive disorder (OCD) who have difficulties controlling their urge to engage in compulsive behaviors. However, unlike the compulsions in OCD, it seems that most individuals with pica do not consume the substance in order to reduce anxiety or stress, or prevent a feared event, but rather describe it as an addiction or a pleasure. Phrases like "I personally love starch" (Young, 2011, p. 12) have been found in Yahoo! discussion groups or other forums. There are, however, case reports of pica eating as compulsions, sometimes reacting to intrusive, ego-dystonic obsessions (see "Comorbidities").

Etiology

No integrative etiological model of pica exists in either children or adults. Over the course of the history of the disorder, various theories emerged, some of which will be explored in detail below.

First, potential etiological factors inherent to the individual, such as personality factors, developmental delay patterns, and traumatic childhood development, are discussed. Afterward, three theories that have been posited comprising social or cultural factors leading to pica, particularly focusing on geophagy, are focused upon.

There are different models comprising the etiology of eating disorders in general, which conceptually include pica (Carter, Wheeler, & Mayton, 2004). Marchi and Cohen (1990) suggested lack of self-control as an underlying factor for pica, associating picky eating with self-control in children on the other hand. In children, especially with intellectual or developmental disorders, pica is sometimes viewed as a simple developmental retardation. It is possible that the behavior of oral investigation and mouthing, being developmentally appropriate until the age of two, could expand over a longer period of time in some children including the ingestion of items that are consistent with pica. Other variables associated with pica in children include potentially traumatic events such as parental separation, neglect, or too little interaction between parents and the child (Singhi, Singhi, & Adwani, 1981). In individuals with intellectual disabilities, pica has been significantly associated with lack of support and social contact in- and outside of the family as well as the absence of involvement (activities or day programs), but has not been associated with interpersonal conflict (Ashworth et al., 2009). These aspects might also be relevant in children without intellectual or developmental disabilities.

In her book on pica, particularly geophagy, Young (2011) posits three main etiological theories of pica:

1. *Earth or other pica substances as a famine food*. There are many accounts of people engaging in geophagy in times of food shortage. There are also incidents in modern history: in 2008, during a food crisis in Haiti, some inhabitants would eat "cookies made of dirt, salt, and vegetable shortening" (Katz, 2008). Populations with the highest pica prevalence rates seem to be in Third World countries or in areas with low socioeconomic status (see section "Epidemiology"). However, this does not explain cases of individuals with pica like Alexia, who do not lack food and do not need the earth for its (questionable) nutritional value. Young's hypothesis (Young, 2011) also does not hold up for the many individuals with pica who consume items that do not hold any nutritional value. Additionally, hunger is likely not a core motivator in all types of pica, as many individuals with pica stick to one specific substance regardless of item availability or their hunger state. In order to test if the hypothesis is at least partially true, Young (2011) performed a review of geophagy literature where data on hunger status of the individuals was included, with some of the reports dating back to the nineteenth century. Only half of these reports found any relation between geophagy and hunger (28% sometimes, 22% always), while the other half stated that the consumption of earth was never associated with hunger and reported geophagy in populations where there was never a supply shortage. In a more recent study (Young et al., 2010), the participants were specifically asked for possible motivations for pica, including hunger. None of the participants gave hunger as a reason for their pica, and indirect indicators of possibly low socioeconomic status (which could prompt hunger that might not be reported directly due to embarrassment) did not differ between individuals suffering from pica and those who did not. In sum, the theory does not explain all cases of pica and pica substances.
2. *Pica as a dietary supplement, mainly because of a lack of micronutrients*. Micronutrient deficiencies related to pica are primarily iron deficiency and to a lesser extent calcium and zinc. This theory is useful to explain the pica prevalence rates in pregnant women who may become deficient in certain micronutrients, particularly iron. Furthermore, there is evidence from case studies that connect pica to micronutrient deficiencies. For example, Young et al. (2010) found a strong correlation between iron deficiency and pica, even after controlling for confounding factors. Interestingly, one could reason that once any micronutrient deficiency was eliminated in individuals with pica, their

pica behavior would remit. While there are many historical accounts for this theory, mainly involving the correction of anemia (e.g., Carlander, 1959), one recent randomized controlled trial (RCT) study investigating this hypothesis in Zambian children (Nchito et al., 2004) reported no decrease in pica behaviors after supplementing iron or multivitamin supplements. In sum, evidence for this particular theory is inconsistent, and, importantly, there are no studies to date allowing for the attribution of causal effects.

3. *Pica as a protection and a detoxifier*. Young (2011) finally posits the theory that in some cases, like geophagy, pica may help to shield individuals from harmful substances or detox if the harmful substances have already been ingested. This theory might explain why individuals with low immune defense, like children and pregnant women, are affected by pica. Bhatia and Kaur (2014b) mention antidiarrheal properties of certain types of clay (e.g., Vermeer & Ferrell, 1985). However, to our knowledge medicinal or therapeutic properties have not been found for other common pica substances besides clay. This also does not explain the ingestion of harmful objects that often leads to medical complications in individuals suffering from pica (see "Associated Impairments").

In sum, the understanding of pica is very limited currently. No model is able to explain the occurrence and maintenance of pica in all subgroups or for all pica substances. More research needs to clarify whether a unified theory exists; how multifactorial models including socioeconomic, biological, and psychological factors should be composited; and how to delineate successful treatment strategies.

Diagnostics and Treatment

Before starting psychological treatment, it is essential to determine how the individuals' symptom presentation fits into a diagnostic classification and select treatment strategies accordingly. The Eating Disorder Assessment for DSM-5 (EDA-5; Sysko et al., 2015) allows for the assessment of pica according to the new DSM criteria. To our knowledge, an adaptation for children is planned for. In areas of the world where pica might be stigmatized, the interview strategies should be chosen accordingly. For example, in the diagnostic process, several variables might be considered, like matching interviewers to the sex and social and ethnic background of the interviewee. To minimize underreporting, normalizing the behavior at the beginning of the interview and using certain rhetoric techniques might be helpful (Young, 2011).

Alternative methods to assess pica behavior in individuals that do not, cannot, or do not want to provide information in self-report are available even if not standardized. Individuals with potential pica can be presented with various nonfood or placebo items for dangerous substances, to assess choices of pica substances (Study III in Foxx & Martin, 1975; Piazza et al., 1998). X-rays can detect potential items in the digestive system (e.g., Anderson, Akmal, & Kittur, 1991). In behavior functional analyses (Hirsch & Myles, 1996; Piazza, Hanley, Blakeley-Smith, & Kinsman, 2000), particular reinforcements for pica behavior instead of pica behavior itself can be analyzed. Admittedly, the latter needs at least minimal engagement of the patient.

Literature on psychological treatment outcome and recommendations is scarce. There are no randomized controlled trials for treatments of pica, but various case studies provide suggestions of different approaches. As many cases of pica show spontaneous remission, especially in children, most reports of treatment attempts are for individuals with intellectual disabilities. However, most of the strategies from these reports can be applied to otherwise healthy youth with pica.

Bell and Stein (1992) concluded in their review that pica "may be a learned behavior subject to change by manipulating antecedent or consequent events in the subject's environment" (p. 378). This has important implications for treatment conceptualization, and as a consequence, most strategies found in case studies can be characterized as behavior modification, e.g., stimulus control, response blocking, and positive reinforcement with pleasant consequences. These and other strategies mainly employed historically

or today in very severe cases are explored in more detail below.

Stimulus control is a traditional strategy used to control or inhibit a behavior. It describes the manipulation of overt or covert behavior through the intended exploration, avoidance, or change of stimuli that precede the targeted behavior (for an example of stimulus control in treatment of an individual with pica, see Piazza, Hanley, & Fisher, 1996). In the case of pica, stimulus control might include the limitation of access to the substance or to the location where the substance is usually consumed. *Response blocking* is often used in individuals with developmental disabilities (Matson et al., 2013), interrupting attempts to engage in pica, sometimes in baiting situations to increase contingency of the treatment. Reportedly, response blocking was used effectively in three individuals with intellectual disabilities (McCord, Grosser, Iwata, & Powers, 2005) and in two other studies, which combined response blocking with alternative real foods (Hagopian & Adelinis, 2001; Piazza et al., 1996). *Positive reinforcement with pleasant consequences* for appropriate behavior is another method used. Differential reinforcement has been given in various ways, but with mixed results (Bell & Stein, 1992). Such reinforcements have included contingency management on reduction of pica, or for behaviors incompatible with pica, mostly stimulating the same senses as the pica behavior using consumable reinforcements. Discrimination training has also been used in children, reinforcing correct answers to questions concerning the problematic behavior (e.g., which substances should or should not be eaten), yielding similarly mixed results, possibly due to differing language functioning (Bell & Stein, 1992).

Besides currently used strategies, some techniques have more of a historical value or are only used in very severe cases, such as significant intellectual impairment or use of highly dangerous substances. There are some accounts of contingently applied *overcorrection* as a treatment for pica, including extensive oral and personal hygiene or tidying of the area pica behavior was typically engaged in (Singh & Winton, 1984). These procedures produced mixed results, but especially the application of oral hygiene seems to be somewhat effective as a treatment (Bell & Stein, 1992). According to Matson et al. (2013), overcorrection has not been used for three decades. There are also accounts relying on contingent *positive reinforcement with unpleasant consequences* for pica behavior. For example, Paisley and Whitney (1989) conducted a series of experiments in an adolescent with intellectual disabilities, including the contingent application of lemon juice in the subject's mouth, reporting decrease of pica. Like overcorrection, this procedure is not socially acceptable today and has more of a historical value (Matson et al. 2013). Other examples of punishment methods include time-outs, like facial screening (e.g., therapist's hand over the eyes of the individual as reported by Fisher et al. (1994) and Falcomata, Roane, and Pabico (2007)). This was shown to be effective in three children in combination with rewarding procedures or enriched environment (see below), respectively. Physical restraint has also been used in case studies, proving to be effective in most (Bell & Stein, 1992). For example, Singh and Bakker (1984) compared an overcorrection procedure to a brief restraint, removing the pica substance and holding the subject's arms. Both were similarly effective, but the restraint was less time-consuming. Hand-held restraint also significantly reduced pica in a woman with intellectual disabilities (Bogart, Piersel, & Gross, 1995). Restraint is controversial, but still used today (Matson et al., 2013). In cases in which the behavior is life-threatening or the person is too intellectually impaired, *self-protection devices* like helmets or restriction jackets have been used (e.g., Rojahn, Schroeder, & Mulick, 1980). This is mostly seen in institutionalized individuals and might reduce pica, but probably increases other maladaptive behaviors, such as stereotypy (Rojahan et al., 1980) and other negative consequences, like reduced social interaction.

Various comorbid disorders or conditions in which pica behavior might occur without warranting a separate diagnosis (see "Comorbidity and Impairment") may warrant treatment first of the other diagnosis and then pica secondly, as pica may remit with the treatment of the primary disorder.

Moreover, psychological issues presenting in the families of the children suffering from pica might need attention prior to engage in the treatment of pica itself (Bhatia & Kaur, 2014b). Besides psychological treatment, *medical treatments* may also be indicated, such as treating potential underlying nutritional deficiencies with supplements. So far, however, nutritional supplementation has not proven effective by an RCT to date (see "Etiology"). SSRIs may be effective in some cases of pica (e.g., Beck & Frohberg, 2005; Bhatia & Gupta, 2009; Bhatia & Kaur, 2014a; Stein et al., 1996) and should be researched further to investigate generalizability of findings beyond treatment of potential underlying symptoms of depression. Surgery represents the most common treatment of associated medical consequences (discussed in "Associated Impairments") (Decker, 1993).

In summary, there are many case studies investigating different treatments, but none (except for micronutrient supplementation) has been tested in an RCT. Existing case studies suffer per definition from missing control groups, if not additional, other major methodological flaws (Bell & Stein, 1992). Due to the different characteristics of populations presenting with pica, presumably various treatment strategies will be necessary to treat pica differentially dependent on the substance consumed. As Matson et al. (2013) put it: "more research is needed investigating algorithms with pica, based on type of behavior, frequency, intensity, chronicity and cause" (p. 2569).

Summary and Outlook

Pica is a disorder that appears to mainly occur in children, pregnant women, and individuals with intellectual disabilities. However, representative epidemiological data is rare and, for the general population, nonexistent. It is characterized by eating substances that are considered nonfood and nonnutritive. It can occur within the context of other mental disorders such as neurodevelopmental disorders, schizophrenia, eating disorders, non-suicidal self-injury, and factitious disorder and is not diagnosed separately unless needing additional medical attention or if not fully explained by the other disorder (e.g., not only eating a nonnutritive, nonfood substance to substitute for real food but also for other reasons in the case of eating disorders). In some cases, pica may be mistaken for a substance use disorder or OCD. Pica is also associated with severe medical impairments (e.g., intestinal perforation or obstruction, infections, or asphyxiation), while little is known about associated psychological impairment. The extremely relevant topic of developmental challenges associated with pica has not been researched to date, beyond our current understanding of potential factors impacting developmental delay, such as shame, withdrawal, depression, and medical hospitalization. Currently, there is no unified etiological model of pica; instead, single theories exist that explain pica behavior with a limited number of substances and/or subgroups. Psychological treatment in children is limited to techniques of behavior modification that are only supported by case studies. In case of pica behavior as part of another mental disorder, the latter should receive treatment first, possibly also leading to a remission of pica behavior. Pharmacological treatment of individuals with pica is scarce, but there is some limited support for the use of SSRIs. And, of course, medical complications should be treated immediately, in most cases needing surgical procedures.

Apart from television shows that feature individuals with pica as a popularized anomaly or weird addiction, pica has not received much attention in the general community. Importantly, general knowledge of pica is scarce in the medical and mental health professional community. In order to inform the public as well as health-care providers, more research is needed to elucidate prevalence rates across populations as well as the longitudinal course of pica. Moreover, experimental studies will help to elucidate etiological mechanisms. Current approaches based on the Research Domain Criteria (RDoC) might be useful to investigate underlying neurological, genetic, and behavioral mechanisms making use of multiple methods investigating the different domains (cognition, social processes, arousal/regulator systems, and negative as well as positive valence

systems) (for an overview over RDoC, see, e.g., Morris & Cuthbert, 2012). Furthermore, future research could utilize models from phenotypically related disorders (e.g., impulse control disorder, substance abuse). These findings will then allow us to develop treatment strategies. Eventually, new treatment strategies can be developed and tested in larger scale randomized controlled trials.

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Anorexia Nervosa and Bulimia Nervosa in Children and Adolescents

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Introduction

Anorexia nervosa (AN) and bulimia nervosa (BN) are serious psychiatric illnesses that often first appear in adolescence. These eating disorders are driven by body shape and weight concerns, are associated with a number of serious medical and psychiatric features, and have mortality rates among the highest of all psychiatric illnesses (Sullivan, 1995). It is important to identify them quickly, as these illnesses worsen over time and are most successfully treated soon after the onset of symptoms.

The diagnostic criteria for AN and BN set forth in the *Diagnostic and Statistical Manual for Mental Disorders*, fifth edition (DSM-5) (American Psychiatric Association [APA], 2013), have undergone recent revisions that have particular relevance for children and adolescents. In particular, the revisions clarify and slightly broaden the diagnoses, to ensure that more patients who present with serious disturbances in eating behavior are properly diagnosed. Many of the changes place more reliance on clinical judgment for accurate diagnosis, making it even more important for clinicians who treat vulnerable populations to be informed about these illnesses. This chapter will describe AN and BN in detail and focus on the manifestations of these conditions, how they are diagnosed, and how they should be treated in children and adolescents.

Presentation of the Current Diagnostic Criteria in DSM-5

Over the course of a year, approximately 0.4% of the population carries a diagnosis of AN, and 1–2% of the population has a diagnosis of BN. Both of these illnesses disproportionately affect children, adolescents, and young adults (APA, 2013). AN and BN affect females more commonly than males, but males account for significant numbers of individuals with eating disorders among pediatric samples. Groups considered at high risk include those with a family history of eating disorders and those for whom low weight is encouraged (e.g., runners, ballet dancers, and wrestlers). While eating disorders present with higher frequency in Western countries and cultures that emphasize thinness, lower rates elsewhere may be partly due to lower likelihood to present for treatment (Gordon, 2000). Further, across all cultures, AN and BN may be under-identified in

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males and ethnic minorities, because these populations have been traditionally considered less likely to develop the disorders.

Anorexia Nervosa

AN is defined as an inability to maintain a healthy body weight (for age, sex, developmental trajectory, or physical health), together with an intense fear of weight gain or continual behavior that impedes it. Patients with AN have a disturbed perception of body weight and/or shape, and these factors overly influence their self-evaluation. They may or may not recognize the severity of their illness. There are two clinical subtypes of AN: binge/purge subtype (AN-BP) characterizes those who experience recurrent episodes of binging and/or purging within the last 3 months and restricting subtype characterizes those whose eating behaviors include restrictive eating exclusively. (APA, 2013)

Body mass index (BMI) is an indication of clinical severity of AN, and, according to DSM-5, guidelines for adults are mild (≥17 kg/m^2), moderate (16–16.99 kg/m^2), severe (15–15.99 kg/m^2), and extreme (<15 kg/m^2). When evaluating children and adolescents, clinicians must evaluate weight in the context of developmental trajectory as well as BMI-for-age percentile. In order to accurately gauge the severity of the condition in these populations, it is important to consider these measurements in the contexts of body type, weight history, physiological disturbances, and functional impairment. Individuals are considered to be in partial remission when they have consistently remained at a healthy body weight but remain fearful of weight gain, continue to engage in restrictive food behaviors, or experience disturbance of their body shape or weight.

Bulimia Nervosa

BN is defined by episodes of binge eating in which an individual loses control over eating and consumes an objectively large amount of food and then compensates by engaging in behaviors such as vomiting, consumption of medications (e.g., laxatives or diuretics), fasting, or doing excessive exercises that aim to prevent weight gain (APA, 2013). To meet criteria for a diagnosis of BN, an individual must not have a current diagnosis of AN, the above-described behaviors must occur at least once a week for 3 months, and shape/weight must exert excessive influence on self-evaluation.

A binge episode consists of an unusually large amount of food eaten during a discrete period of time. The DSM-5 allows for clinical judgment to determine what meets this standard and specifies that context is important (e.g., if others are eating similarly large amounts, such as what often occurs at celebrations, a considerable quantity of food may be reasonable). A limited period of time can broadly be considered as less than 2 h, although again, clinical discretion applies. The binge episode may be planned or unplanned and may occur in one or multiple settings, but is characterized by a continuous sense of loss of control. Often individuals with BN eat secretly, consume food during a binge they would otherwise avoid, and continue eating until they feel uncomfortably full.

Loss of control is defined subjectively and may be described as not being able to stop eating or control the amount of food consumed. Some patients describe binge eating as being similar to a ball rolling down a hill, reporting that once they begin a binge, they cannot stop until it comes to its natural end point. Functional impairment associated with binge eating varies: some report that they cannot stop binge eating in order to answer the phone, but can do so at the unexpected presence of a roommate or family member.

Clinical severity of BN is indicated primarily by the average frequency of inappropriate compensatory behaviors per week. Guidelines provided in DSM-5 are mild (1–3 episodes), moderate (4–7 episodes), severe (8–13 episodes), and extreme (>14 episodes). As with AN, features such as functional impairment caused by BN should also be considered when determining severity. Individuals are in partial remission when some, but not all, criteria are met for a prolonged period of time.

History of Eating Disorder Diagnoses

Clinical descriptions of an AN-like syndrome have been present since the 1600s, and the illness received its current name in the late 1800s (Bell, 1985). AN appeared in the first edition of DSM (1952) as a psychophysiological reaction (APA, 1952) and in DSM-II (1968) under "special symptoms – feeding disturbances" (APA, 1968). Its portrayal in DSM-III (1980) was the first to resemble its current depiction: criteria included low body weight, fear of weight gain, and disturbed body image (APA, 1980). DSM-IV (1994) was the first to describe subtypes for AN and included a system of diagnostic priorities in which patients could not receive simultaneous diagnoses of both AN and BN.

BN was formally recognized more recently than AN, described as a variant of anorexia nervosa in a 1979 case series by Russell (Russel, 1979), and appearing in DSM-III as a condition of recurrent binge eating episodes followed by distressing thoughts. In DSM-III, BN was characterized by any three of the following: binge eating on high-calorie foods; secret binging; termination of a binge episode because of sleep, abdominal pain, social interruption, or self-induced vomiting; repeated attempts to restrict, vomit, and use cathartics or diuretics; and frequent weight fluctuations. DSM-III-R (1987) was the first version to define BN as recurrent episodes of binging and purging (APA, 1987), and in DSM-IV, a more current definition of BN took hold (APA, 2000).

DSM-5 includes several small but important changes regarding eating disorders. The language has a stronger behavioral focus than that of DSM-IV, and DSM-5 criteria require decreased frequency and severity of symptoms.

Many of the problems with previous diagnoses of AN and BN stemmed from the rigid application of the diagnostic criteria included in DSM-IV, which left many individuals to receive the nonspecific diagnosis eating disorder not otherwise specified (EDNOS). Adolescents were significantly affected: as many as 80% of those with an eating disorder were diagnosed with EDNOS according to DSM-IV criteria (Le Grange, Swanson, Crow, & Merikangas, 2012). EDNOS had limited clinical utility; its heterogeneous nature increased barriers to specialized care and made research to understand and treat these illnesses more challenging.

The DSM-5 aims to reduce the numbers of clinically affected individuals who received the nonspecific EDNOS diagnosis and improve the clinical utility of the diagnostic labels for psychiatric disorders (Call, Walsh, & Attia, 2013; Ornstein et al., 2013). By better clarifying the diagnostic criteria, and decreasing some of the symptom frequency required for meeting diagnostic threshold, DSM-5 increases the likelihood that patients receive accurate and specific diagnoses. In fact, in two epidemiological studies of 3048 female high school and college students, there were 118 cases of eating disorders, of which 86 (72.9%) were diagnosed with EDNOS according to the DSM-IV criteria. Use of the DSM-5 criteria decreased the number of individuals who received this residual diagnosis to 52 (44%), with commensurate increases in those receiving AN and BN diagnoses (as well as a small group receiving a diagnosis of avoidant/restrictive food intake disorder (ARFID), a newly clarified diagnosis that describes restrictive eating that is not associated with body weight or shape concerns) (Machado, Gonçalves, & Hoek, 2013). Importantly, the symptoms of these patients, who would previously have been diagnosed with EDNOS and are now receiving formal diagnoses according to DSM-5, are no less clinically severe than those who met criteria for AN and BN according to the more stringent DSM-IV criteria (Le Grange et al., 2013).

Anorexia Nervosa

DSM-5 preserves the core features of AN as defined in DSM-IV, including low body weight and body shape or weight disturbance, but clarifies the wording of the criteria (APA, 2013, 2000). It describes significantly low body weight as being low in the context of age, sex, developmental trajectory, and physical health, and it

eliminates the commonly misinterpreted example that was included in the low-weight criterion in DSM-IV (e.g., <85% expected). DSM-5 describes low weight as less than minimally normal for a patient's height and age. The accompanying text offers guidelines to define low weight, such as the use of BMI for adults and BMI-for-age percentile for children and adolescents, and allows for clinical judgment regarding other factors like body build, weight history, and physiological disturbances. This clarification is especially important for children and young adults, for whom individual context is vital in determining the severity of low weight.

DSM-5 criteria no longer require that a patient with AN exhibit or endorse fat phobia. Instead, the presence of behavior that interferes with weight gain is included as part of criterion B. Individuals who exhibit persistent behavior that prevents weight gain meet the criteria for diagnosis regardless of their ability to articulate why they are engaging in these behaviors. This change is particularly important for children and adolescents whose abstract reasoning skills may not yet be fully developed or who may have more difficulty understanding and expressing the cognitions associated with their behaviors (Ornstein et al., 2013).

Finally, DSM-5 eliminates the requirement of amenorrhea in post-menarchal women. The modification is consistent with DSM-IV criterion exceptions that allowed women who take exogenous hormones, adolescents who have not yet reached menarche, and men to meet criteria. The change was made because menstrual status has been found to be a poor indicator of clinical severity (Attia & Roberto, 2009).

Bulimia Nervosa

As with AN, DSM-5 preserves the core features of DSM-IV's BN diagnosis, including disturbance of body shape or weight and recurrent episodes of binge eating and purging. However, because of data about course and prognosis among those with binge and purge behaviors at different frequencies (Trace et al., 2012), DSM-5 reduces the frequency of episodes required for a diagnosis of BN from twice to once weekly for a 3-month period. These criteria help decrease the numbers of individuals who receive a nonspecific diagnosis (other specified feeding and eating disorders (OSFED)) and improve the description of the individuals whose regular pattern of binge eating and compensatory behaviors requires clinical attention (APA, 2013).

As a result of this modification, individuals who have begun engaging in such behaviors may be recognized sooner and treated earlier, before the presentation of more frequent symptoms with associated physical and/or functional impairment. As with AN, this is particularly significant for children and adolescents who have begun demonstrating behavioral criteria, but have not yet developed more severe symptoms. Earlier identification invites immediate targeted treatment and increases the chance of successful intervention.

DSM-5 also removes the differentiation that DSM-IV included of two distinct (purging and non-purging) subtypes of BN, as the subtypes were not considered clinically useful. Clinicians rarely used these subtypes; also individuals with BN respond similarly to treatment regardless of their method of purging (Wolfe, Hannon-Engel, & Mitchell, 2012).

Etiology

Multiple factors, including genetic and environmental influences, contribute to eating disorders. While the specific etiology of AN and BN is still unknown, clinicians have found it useful to look at predisposing and precipitating factors separately. Genetics unequivocally play a role in the development of AN and BN. The heritability of both illnesses is moderately large, as is indicated by higher rates in identical twins than in fraternal twins (Bulik et al., 2006). Linkage analysis and studies on serotonergic and dopaminergic pathways, as well as disturbances in the central nervous system, additionally indicate a significant biological contribution to these illnesses (Pinheiro, Root, & Bulik, 2009). Research to

further understand these genetic contributions and how they operate is a major area of exploration in the field.

Epigenetic changes to gene expression may also serve as precipitating factors for AN. These gene-environment interactions may involve changes in the in utero environment that occur as a function of maternal stress, nutrition, or other factors (Bulik et al., 1999). Women with AN who are able to conceive often have minimal weight gain during pregnancy.

Adolescents' hormonal changes, growth trajectory, and common pursuit of achieving social normalcy, including idealized form, may also play a role in the development of eating disorders. For individuals with a genetic predisposition, a developmental milestone such as pubertal maturation or leaving home may contribute to the onset of an eating disorder (APA, 2013).

A number of temperamental factors have additionally been identified as risk factors for both illnesses. Those diagnosed with AN and BN often have high levels of anxiety, depression, obsessive-compulsive symptoms, and interpersonal sensitivity (Casper, Hedeker, & McClough, 1992). Patients with AN are often self-disciplined and conscientious. Their cognitive styles are often inflexible, they may be less adventurous than average, and they frequently score higher on a reward dependence scale (Bulik, Sullivan, Weltzin, & Kaye, 1995). Patients with BN are often impulsive and may have a more dramatic temperament profile, exhibiting traits such as exploratory excitability and extravagance. Many of these personality traits are elevated in unaffected relatives of patients as compared to unaffected controls, suggesting that the traits themselves are hereditary (Wade et al., 2008).

Environment also plays a significant role in development of these disorders. Societal pressures and unrealistic expectations about thinness, beauty, and achievement serve as precipitating factors for both BN and AN, and Western culture's idealized images of beauty contribute to disordered eating and interest in weight loss (Becker, Burwell, Gilman, Herzog, & Hamburg, 2002). Occupations such as athletics and modeling, which emphasize thin physique, are associated with higher rates of eating disorders (APA, 2013). Further, family members who have disordered eating may expose children at an early age to these patterns of behaviors.

Traditional views of the causes of AN and BN included the theory that inappropriate familial relationships contribute to the development of these illnesses (Minuchin et al., 1975). Data do not support the association between particular family disturbances and the development of these conditions. In fact, the empirically supported family-based therapy, often called the Maudsley method, specifically includes discussions with family members about this lack of evidence, insisting that families are "not to blame" for eating disorders (Lock et al., 2010; Loeb et al., 2007). Similarly, while some have proposed sexual abuse and other trauma history as risk factors, there is little support for these being unique predictors for eating disorders. History of sexual abuse is more common among individuals with psychiatric diagnoses than it is among the general population, but rates of past trauma are not higher among those with eating disorders than those with other psychiatric illnesses (Welch & Fairburn, 1994).

Symptom Presentation

Often, eating disorders are not identified immediately; clinicians must ask informed questions about eating and associated behaviors in order to successfully diagnose them (APA, 2013; Walsh, 2008). Even in the face of significantly disordered eating, medical symptoms may be minimal, and laboratory assessments may be normal. Patients may engage in problematic eating behaviors in private or wear loose-fitting clothing to conceal their weight loss. Children and adolescents with AN and BN often deny their symptoms of illness, rarely seek help for weight loss, and may have few physical complaints. When they do present to pediatricians or subspecialists, patients frequently express concern with the somatic or psychological sequelae of starvation (e.g., mood changes, bloating, abdominal discomfort, or fatigue) rather than the disordered

symptoms themselves. The illnesses may be noticed first by friends, teachers, or siblings, rather than parents or physicians.

Younger patients with atypical or more subtle presentations of the disorders are at high risk for delayed or missed diagnoses. This is especially true for those who are not fully grown and may maintain or gain weight (e.g., less than expected) despite a diagnosis of AN and those who are overweight prior to the development of the disorder.

Many of the physical manifestations of eating disorders result from malnourishment or compensatory behaviors aimed at preventing weight gain. Although certain physiological complications may be more subtle in BN than AN, patients who purge, regardless of their diagnosis, have significant overlap in symptoms and may manifest physiological disturbances in all major organ systems. Many of these effects are reversible with weight restoration and cessation of compensatory behaviors.

Initial Presentation

Patients with AN may have unexplained weight loss, failure to make expected weight targets or growth milestones, significant fluctuations in weight, or delays in pubertal development. They may complain of feeling cold, having dry skin, thinning scalp hair, and developing fine hair on their face, arms, or trunk (i.e., lanugo). Facial wasting and cachexia may be observed. All of these symptoms are secondary to malnutrition.

Patients with BN are normal or above normal weight, but may experience large weight fluctuations. They (and patients with AN-BP) may have swollen parotid glands (common immediately following cessation of purging symptoms) and/or Russell's sign (scarring calluses on the dorsal aspect of their hands as a result of scraping teeth against their skin while inducing vomiting). They may have dental caries or erosion of tooth enamel, which is often discovered by a dentist rather than a pediatrician. Some patients may complain of a sore throat, a result of frequent vomiting.

Mood and Behavioral Symptoms

Patients with eating disorders often become increasingly obsessive (particularly about food), may experience shame and guilt about eating, and often eat in secret. For many, self-worth becomes intricately tied to body weight. They view weight loss as an achievement and sign of willpower and weight gain as an intolerable failure. They may impose strict rules about their intake or exercise regime and be preoccupied with thoughts about food, shape, and weight to the point of functional impairment. Patients may weigh themselves frequently and vigilantly monitor their body shape through techniques like measuring or pinching themselves. Many attribute these rigid behaviors to a sense of maintaining control. Some become functionally impaired and begin to avoid work, school, or social functions. Patients commonly deny the severity of their condition, although this is not necessarily pathological in younger patients, for whom displaying deficits in risk assessment is developmentally appropriate (Boyer, 2006).

Depression and obsessionality are both caused and exacerbated by a starved state (Keys, Brozek, Henschel, Mickelsen, & Taylor, 1950). This was demonstrated in a study by Keys and colleagues, in which healthy men who were conscientious objectors during World War II voluntarily decreased weight to 75% of their pre-starvation body weight and, in a malnourished state, began to exhibit cognitive impairments, depression, and preoccupation with food. Social isolation, insomnia, irritability, decreased libido, and unusual eating rituals, symptoms frequently described among AN patients, were also seen in the malnourished experimental sample described by Keys et al.

Patients with BN may experience depression and anxiety as well as other comorbid conditions. Binge eating may be triggered by a variety of factors aside from hunger cues, including interpersonal stressors, negative affect (particularly relating to food, shape, and weight), and boredom. These episodes often take place in the late afternoon or evening, following a period of dietary control in the early part of the day, and may contain sizeable caloric content (some

research suggests an average of greater than 3000 kilocalories) with a high macronutrient content of fats and carbohydrates (Walsh, Hadigan, Kissileff, & LaChaussée, 1992). Patients often report feeling numb during binges and intense shame and regret following their completion.

Outside of binge episodes, patients with BN often heavily restrict food intake and attempt to follow rigid rules about eating. These patterns set the stage for intense food craving and for a feeling of failure when identified food rules have not been obeyed. Episodes of binge eating and purging frequently follow. The post-purging nutritional depletion may contribute to hunger that sets the stage for additional binge eating episodes. Physiological disturbances that affect satiety (Kissileff et al., 1996), such as enlarged stomach capacity (Geliebter et al., 1992), delay in stomach emptying, and reduction in release of cholecystokinin (a peptide hormone that drives cessation of eating) (Devlin et al., 1997), further perpetuate these factors and the binge-purge cycle. Notably, a patient's decision to binge seems to have a significant effect on his/her sensation of satiety (Zimmerli, Devlin, Kissileff, & Walsh, 2010), and the prokinetic agent erythromycin, while accelerating gastric emptying in patients with BN, does not decrease binge behaviors (Devlin et al., 2012). For this reason many clinicians are suspicious of the role of gastrointestinal abnormalities in the persistence of BN (Heaner & Walsh, 2013).

Patients purge in a variety of ways, including stimulating their gag reflex (using fingers and tools or even at will); taking laxatives, diuretics, or enemas; misusing medications (usually thyroid hormones, insulin, or diet pills); and doing driven exercise (when patients exercise excessively, often interfering with other aspects of their life and health) (APA, 2013).

After purging, individuals report feeling physical relief from uncomfortable fullness as well as mitigated fear of gaining weight. Physical and psychological relief resulting from purging is temporary, however, and long-term purging causes irregular hunger cues, lowered self-value, and a continuation of the binge-purge cycle. Moreover, for some individuals, the behavioral cycle becomes so engrained that binge eating becomes a means to purging and the short-term relief that results.

Gastrointestinal System

Gastrointestinal symptoms are among the most commonly reported physical symptoms among individuals with eating disorders. Patients frequently report bloating, nausea, constipation, and postprandial fullness. Malnutrition in AN can lead to delayed gastric emptying, constipation, hepatic transaminitis, and dyslipidemia. Gastrointestinal abnormalities in BN are more subtle, but include enlarged gastric capacity and delayed gastric emptying abilities. Vomiting may be associated with esophagitis, gastroesophageal reflux, hematemesis, and parotid swelling. Rarely, esophageal tears (i.e., Mallory-Weiss tears) occur in patients who vomit, gastric rupture in those who binge severely, or rectal prolapse in those who abuse laxatives (APA, 2013). Occasionally a foreign body used to purge (e.g., a utensil) is accidentally swallowed and has to be removed (Ruiz & Soler-Gonzalez, 2005).

Cardiovascular System

AN and BN patients may present with headaches, syncope, and exercise intolerance. Common physical examination findings among these patients include bradycardia, hypothermia, and hypotension, sometimes associated with complaints of dizziness or faintness. Electrolyte abnormalities, such as hypokalemia, may be seen in patients who purge regularly, and hyponatremia may be seen among those who consume large amounts of water. Hypophosphatemia and hypomagnesemia may be present in those who are significantly nutritionally depleted and may be markers for refeeding syndrome. Cardiac arrhythmias are possible, including QTc prolongation. Patients with AN and BN may have orthostatic changes in pulse and blood pressure due to dehydration. Additionally, patients with AN are at risk for mitral valve prolapse and pericardial fusions. Abuse of ipecac, sometimes used for

purging, can lead to nerve and muscle damage including cardiomyopathy.

Endocrine System

Malnutrition and compensatory behaviors aimed at preventing weight gain may lead to suppression of hormone levels resulting in pubertal delay and/or menstrual irregularities. Post-menarchal females with AN often have amenorrhea, and those with BN may have amenorrhea or oligomenorrhea. Younger patients are at a great risk for loss of bone mineral density and osteoporosis. Additionally, reduced levels of thyroxine and triiodothyronine can affect thyroid function. Patients may have increased cortisol and lower levels of follicle-stimulating hormone, estradiol, and testosterone.

Renal System

Patients with eating disorders may become dehydrated as a result of poor fluid intake, vomiting, and laxative or diuretic abuse. Renal insufficiency is rare, but has been reported, in individuals with chronic AN.

Neurologic System

Patients with AN may complain of poor concentration, decision-making, or memory, all symptoms that are secondary to starvation. Neuroimaging indicates significant structural changes including decreased white matter volume and enlarged ventricles among those with AN (Kerem & Katzman, 2003). Further research is needed to determine if these abnormalities fully resolve with weight restoration.

Additional Laboratory Abnormalities

Leukopenia is commonly seen in patients with AN due to bone marrow suppression. Cells appear to function despite low concentrations on blood count assessment. Anemia and thrombocytopenia are also possible. Hypercholesterolemia and elevated serum transaminases are common among these patients.

Compensatory behaviors may lead to additional disturbances. Vomiting may lead to the development of high serum amylase with metabolic alkalosis; laxative abuse may be associated with metabolic acidosis. Diuretic abuse or overall dehydration may lead to hypernatremia, hypochloremia, and poor kidney function. Vitamin deficiencies, most notably decreased levels of iron and vitamin B12, may be observed. Patients may also have increased blood urea nitrogen or creatinine in the context of dehydration.

Refeeding Syndrome

Both physiological and psychological recovery from AN are entirely reliant on weight restoration, but additional physiological complications may develop upon initiation of nutritional rehabilitation because limited stores of needed nutrients (e.g., phosphorus or magnesium) are used for initial catabolism. The risk of refeeding syndrome is directly correlated with severity of low weight. Peripheral edema, hypophosphatemia, hypokalemia, hypomagnesemia, and cardiovascular problems are of particular concern (Mehler, Winkelman, Andersen, & Gaudiani, 2010). Thiamine, vitamin D, and a multivitamin should be administered before feeding starts, and potassium, calcium, phosphate, and magnesium should be regularly checked (Mehanna, Moledina, & Travis, 2008). Additionally, vital signs should be monitored daily during refeeding for weight restoration, to watch for elevated heart rate. Edema should be monitored closely as well. Among severely low-weight patients, caloric intake should begin no higher than 1800 kcal/day and be increased slowly over the first week.

Associated Impairments and Comorbidities

Significant numbers of individuals affected by AN or BN may meet criteria for another eating disorder during their lifetime. For those who migrate

from one eating disorder diagnosis to another, AN often occurs first, and BN, OSFED, or binge eating disorder (BED) develops later (APA, 2013). Moreover, both AN and BN are commonly comorbid with mood disorders (specifically major depressive disorder and dysthymia), anxiety disorders, and substance use disorders (Hudson, Hiripi, Pope, & Kessler, 2007).

Anorexia Nervosa

As described above, anxiety and depression frequently co-occur with AN, and malnutrition is considered contributory to these conditions when they occur during the low-weight state. Among patients diagnosed with AN, depression has a lifetime prevalence of just under 40%, and anxiety disorders (especially obsessive-compulsive disorder and social phobia) have a lifetime prevalence of just under 50% (Hudson et al., 2007). A significant number of patients with AN, 25%, are diagnosed with a substance use disorder at some point in their lives.

Bulimia Nervosa

Among individuals with BN, mood disorders (unipolar depression as well as bipolar disorder) have a lifetime prevalence of just above 70%, and anxiety disorders have a lifetime prevalence of just above 80% (Hudson et al., 2007). These diagnoses are often integrally linked with BN's symptoms—beginning around the same time as the BN behaviors and improving with eating disorder treatment (Attia & Walsh, 2009). Among the anxiety disorders common in BN patients are agoraphobia, specific phobia, generalized anxiety, and post-traumatic stress disorders. More than half of patients with BN meet criteria for personality disorders (e.g., borderline personality disorder) (Bulik, Sullivan, Joyce, & Carter, 1995), and just below 40% of BN patients have a substance use disorder at some point in time. This is consistent with the finding of high levels of impulsivity reported among patients with BN, including behaviors such as stealing and self-harming. Indeed, patients with BN may have a comorbid impulse control disorder, including intermittent explosive disorder, attention-deficit/hyperactivity disorder, or oppositional defiant disorder. The phenomenological resemblance between binge eating behaviors and substance abuse has been noted in the literature, and there may be common pathways and brain mechanisms that link the two disorders to systems of reward; however, no research currently substantiates the claim that physiological addiction plays a role in BN (Walsh, 2008).

Course and Developmental Challenges

AN often first occurs in mid-adolescence and BN in young adulthood; the onset of either condition before puberty or after age 40 is less common (APA, 2013). Due to the young ages of most individuals affected by AN and BN, developmental challenges frequently occur in the course of these illnesses.

Young adults already struggle to determine their identity and values and begin to grapple with relationships of all kinds. They strive for independence yet still rely on their families of origin. They undergo sexual, hormonal, and physical changes, and young women experience an increase in body fat. Girls in particular often feel self-conscious and insecure at this age. These developmental challenges may serve as nonspecific risk factors for eating disorders. In turn, eating problems may contribute to growth and other developmental delays that often require parental involvement during the course and treatment of AN and BN.

Anorexia Nervosa

The initial weight loss that begins the cascade of symptoms that define AN is frequently precipitated by a stressful event (Walsh, 2008). For example, some patients report that symptoms began after they left home for the first time. Others relate onset of AN to a critical comment

about body shape or weight from a clinician or coach. The illness often begins with common dieting behaviors and a small amount of weight loss that is met with positive reinforcement, which may encourage an association between weight loss and a sense of accomplishment. Patients with a fully developed disorder may become depressed and withdrawn. Younger patients, even at this point, may deny starvation or any "fear of fat" and only complain of weakness or other somatic symptoms. While cognitive impairment in such a starved state is not uncommon, some continue to function at a high level in work or school. Often patients cite their success in these areas as support for their "lifestyle," denying the severity of their illness.

Bulimia Nervosa

As with AN, BN may begin with dieting or fasting, and a restrictive pattern may be present for some weeks or months before binge eating or purging behaviors begin (Walsh, 2008). Hunger cues may lead an individual to overeat, and distress from the binge episode may lead to compensatory vomiting. The patient with BN may at first be pleased that he or she is able to eat without weight gain; however, over time, the cycle leads to binges of increased size and frequency. Mood and physiological and environmental triggers worsen, and as problematic behavioral symptoms increase and become more frequent, BN patients develop further decreased self-esteem, rigid eating rules, and preoccupation with shape and weight.

Treatment

The goals of treatment for AN and BN are to restore weight (in AN), normalize eating, and encourage healthier thoughts and feelings about body shape and weight (Walsh, 2008). To accomplish this, treatment may employ behavioral, family, cognitive, and supportive techniques. Severity of symptoms and medical complications are the primary factors used to determine the appropriate level of care (i.e., treatment setting), although other considerations include patient's (or parent's) motivation for or ambivalence about treatment, psychosocial functioning, availability of resources (e.g., insurance or geographic location), and duration of illness (Freidl, Keegan, Richter, Mayer, & Attia, 2014). Because of the severe consequences of illness progression, the threshold for initiating treatment in children and adolescents should be low. Although voluntary treatment is preferable, treatment is often appropriate even when a patient feels partially or fully coerced (Attia & Walsh, 2009).

Requirements for Inpatient Care

Inpatient treatment provides a highly structured, supportive setting to promote effective behavioral changes. Hospitalization may take place in either a pediatric, medical, or psychiatric unit, and doctors may suggest this type of intensive treatment in order to stabilize medical status and begin the course of refeeding and behavioral change. Indications that this highest level of care is appropriate may include severe malnutrition or dehydration, abnormal laboratory or cardiac assessments, precipitous weight loss, failure to make appropriate weight gains, arrested growth, severe physiologic instability, acute food refusal, uncontrollable binge eating and purging, and failed treatment in a less structured setting. Those who weigh lower than 75% of their ideal or recommended body weight are at particularly high risk for medical complications and should consider hospital-based care. Comorbid illness and poor behavioral control may be additional reasons to consider hospital settings for eating disorder treatment.

Because of the short-term nature of hospitalization, its major goals include medical stabilization, the first stages of refeeding and discharge planning. Residential treatment programs are useful for individuals who do not need acute management of medical and psychiatric symptoms, but need full-time supervision of eating and related activities. Several specialized residential programs have been developed for individuals with

eating disorders who wish to participate in several weeks (occasionally months) of supportive care. For some patients, residential treatment serves to extend the intensive treatment that begins in hospital settings.

Outpatient Care

Outpatient treatment is effective for BN and may be the first line of care for less clinically severe patients with AN. Intensive outpatient programs (IOPs) meet several times a week for a few hours, and partial hospitalization programs (PHPs) meet 4–7 days a week for a large part of the day (Freidl et al., 2014). Both IOP and PHP provide regular monitoring and supervised meals for patients who are in need of support and accountability for the disorder or for relapse prevention. They may be useful treatment settings for those who are acutely symptomatic and/or have impaired social, educational, or vocational functioning, and they often serve as a "step-down" for individuals who have completed a program with a higher level of care.

For those not in a structured program, a multidisciplinary treatment team of professionals is often used for clinical management. Teams often include psychiatrists, psychologists, nutritionists, pediatricians, and other health professionals who meet with the patient in an individual or group setting. Often a team of specialists plays an ongoing role in continued care and relapse prevention after patients complete structured programs.

Anorexia Nervosa

There are many obstacles to providing care for patients with AN (Walsh, 2008). Because of the ego-syntonic features of the illness, patients often state that their eating behaviors are "choices" or "decisions" that they have selected rather than symptoms. Even those pursuing recovery are often reluctant to gain weight or change their behaviors. Patients may have a profound mistrust of others, or be overly sensitive to coercion, making it more difficult for clinicians to develop a therapeutic alliance. Clinicians often find common ground with the patient on issues both parties acknowledge to be problematic—low mood, anxiety, social isolation, loss of control over eating, or physical weakness.

Clinicians encourage independent eating and general functioning when possible, while still emphasizing the severity of the illness and importance of challenging eating-disordered behaviors. Evidence points to the effectiveness of structured programs (e.g., hospital-based care, IOPs, PHPs, residential programs, or family-based treatment) for weight restoration in AN (Attia & Walsh, 2009).

Nutritional Rehabilitation

Weight restoration and nutritional rehabilitation are the first and often among the most difficult steps toward recovery in AN (Attia & Walsh, 2009; Crisp et al., 1991; Guarda & Heinberg, 2004). Programs aim to restore patients to a healthy weight range, as well as normalize disordered eating. One pound of weight gain requires 3500 kcal beyond those necessary to maintain weight (approximately 2000 kcal per day). Therefore, after a patient achieves medical stability on an initial caloric prescription (e.g., 1800 kcal/day), programs may begin to gradually increase intake under medical supervision. Structured treatments commonly prescribe upwards of 3000 kcal daily divided into meals and snacks in order to assure steady weight gain. Nutritional supplements may also be used, and recommended weekly weight gain rates usually range from 2 to 5 lbs. (1–2 kg) in highly structured settings, with lower rates of weight gain expected when treatment is less intensive. Patients are reintroduced to their "fear foods" and encouraged to abstain from disordered behaviors such as cutting or abnormally mixing foods.

Meal supervision ensures that patients consume and retain the calories from meals. Supervision is especially necessary following meals and snacks, when patients may have urges to purge or exercise. Vigilance is also important during weigh-ins because patients may try to water-load, wear or hide heavy items, or otherwise distort their true weight.

Behaviorally based methods have been shown to be the most effective ways to refeed patients (Wiseman, Sunday, Klapper, Harris, & Halmi, 2001). Patients are presented with behavioral expectations, such as eating 100% of their food and attaining a specific minimum weight gain rate, and these behaviors are reinforced with program privileges and special activities. When goals are not met, additional supervision or increased calorie prescription (including nasogastric feeding if needed) may be instituted to improve clinical progress. Staff at treatment programs emphasize that interventions are meant to help patients reach agreed-upon treatment goals, not punish them for "bad behavior."

Patients often experience somatic and emotional discomfort with the caloric intake necessary for weight gain and the cessation of disordered behaviors. Patients are asked to consider food as their medicine, and support and encouragement are provided. In an effort to externalize the illness, treatment providers may encourage patients to "choose not to listen to" the illness or may describe the illness as "clever" or even "parasitic," "infecting" the patient's mind and influencing thoughts, feelings, and ability to engage in healthy behaviors in spite of the suffering it inflicts (Loeb, Hirsch, Greif, & Hildebrandt, 2009). Patients often need to be reassured that the rates of weight gain that treatment providers expect them to achieve are not dangerous when medically monitored and that the health risk of remaining in a starved state greatly outweighs the potential risks of refeeding. It should be communicated that nutritional restoration is effective in restoring physical health as well as improving cognitive function and emotional regulation.

Psychotherapy

Psychotherapy alone has been only modestly effective in assisting weight restoration in AN patients (McIntosh et al., 2005). However, behaviorally focused nutritional rehabilitation programs usually incorporate psychotherapy techniques including cognitive behavioral therapy (CBT), dialectical behavioral therapy (DBT), interpersonal therapy (IPT), and family-based treatment (FBT). Psychotherapy addresses the importance of normal eating and weight and may challenge patients' excessive concern about shape and self-evaluation based on weight as well as interpersonal and food-related anxiety. Psychoeducation includes discussion of healthy eating behaviors that will not interfere with social, familial, educational, or professional goals.

CBT, a time-limited treatment that approaches psychological and behavioral problems with techniques rooted in theories about cognition, behavior, and emotion, has a large evidence base for the treatment of eating disorders (Fairburn, 2008; Dalle Grave, Calugi, El Ghoch, Conti, & Fairburn, 2014; Cooper & Stewart, 2008). Its central tenets include Beck's theory of cognition (that negative automatic thoughts are grounded more in self-schemas than reality), learning theory (classical, operant, and observational learning), mindfulness (relationship with thoughts and feelings), and motivational interviewing.

Essential elements of CBT include psychoeducation (e.g., the physiological cycle of binging and purging), self-monitoring (reflection of behaviors and feelings), cognitive restructuring (identification and testing of automatic thoughts and cognitive distortions), and relaxation (e.g., diaphragmatic breathing or progressive muscle relaxation).

Family-Based Treatment

Also known as the Maudsley method because of its introduction by clinicians at London's Maudsley Hospital (Le Grange, Eisler, Dare, & Russell, 1992), FBT is a manualized therapy in which clinicians work with families to encourage parents to refeed their undernourished child. FBT has a large evidence base in children and adolescents with a short (less than 3 years) duration of AN (Lock et al., 2010; Loeb et al., 2007). The treatment is generally offered over 6–12 months and takes places in three phases together comprising 15 to 20 sessions over the course of 1 year (Lock & Le Grange, 2013).

FBT counters the view that parents are responsible for illness development and instead postures that families play an integral role in recovery. FBT can be compared to an intensive outpatient

program that takes place primarily in the home. The treatment is most effective when the whole family is engaged and presents a united front against the illness—parents are empowered to prepare and supervise appropriate meals, and siblings are encouraged to provide emotional support for the ill child in the family.

The primary goal of FBT is to provide the adolescent with the necessary supervision and support to treat the illness with minimal disruption to his or her developmental trajectory. The patient is allowed to maintain autonomy in areas unrelated to the illness and is ultimately expected to regain control in all areas appropriate for the child's age. The treatment effectively restores weight between 65 and 80% of adolescents with AN and has a 5-year follow-up success rate of 75–90% (Eisler et al., 1997). Other notable benefits include its cost-effectiveness and focus on maintaining appropriate developmental trajectory. Its primary limitation is the limited number of qualified providers (Loeb et al., 2009).

In the first phase of FBT, the therapist meets weekly with the family as the parents aim to restore their child's weight to a healthy range. The therapist helps to externalize the illness, emphasizing that neither the parents nor the patient is to blame. To help the adolescent find motivation for recovery, the therapist stresses the serious medical and psychological risks of the illness, and family members discuss their concerns. The therapist assists the parents and aligns with the patient to relay a clear message—that the illness is onerous and recovery difficult, but that starvation is not an option. The patient must normalize weight and eating behaviors in order to get well. All meals are supervised during the first phase of FBT, and goals serve as useful incentives to encourage the patient to challenge him-/herself. At first, the goals might include avoiding hospitalization. Later, the patient's goals might include regaining privileges, such as attending unsupervised meals with friends or rejoining sports teams.

Phase two of treatment begins when the patient starts to eat more independently, has gained weight to a minimally healthy level, and has less conflict with his or her parents about food. Sessions occur biweekly as parents gradually return control of eating to their child. Meanwhile, the therapist continues to discuss issues surrounding symptoms and weight gain as well as day-to-day issues relating to food or physical activity (e.g., socializing or sports). If the rate of weight gain decreases as the adolescent becomes more autonomous and reintroduces exercise during this phase of treatment, this is addressed in these sessions as well.

The aim of phase three of FBT is to help the patient develop an age-appropriate identity. It consists of monthly sessions, beginning when the patient is able to maintain 95% of his or her ideal body weight. Therapy begins to address more general issues of adolescence and focuses specifically on personal autonomy and appropriate parental boundaries. Relapse prevention is discussed to prepare for termination of treatment. The therapist emphasizes the importance of avoiding weight loss, either deliberate or nondeliberate, and discusses methods to manage it should it occur.

Pharmacotherapy

Although antidepressants are often prescribed, there is little evidence suggesting that tricyclic antidepressant medications (Lacey & Crisp, 1980; Biederman et al., 1985) or selective serotonin reuptake inhibitors (SSRIs) (Attia, Haiman, Walsh, & Flater, 1998; Holtkamp et al., 2005) have a significant effect on rate of weight gain, eating disorder symptoms, or depression in underweight or weight-restored patients. Other medications that have been tried but have demonstrated no clinical benefit for treating AN are the antianxiety agent alprazolam (Steinglass, Kaplan, Liu, Wang, & Walsh, 2014), the prokinetic agent cisapride (Stacher et al., 1993), and the appetite-stimulating agent tetrahydrocannabinol (Gross et al., 1983).

Recent evidence suggests that olanzapine, an atypical antipsychotic medication, may be successful at increasing the rate of weight gain and reducing obsessional symptoms in patients with AN (Attia et al., 2011; Brambilla et al., 2007; Bissada, Tasca, Barber, & Bradwejn, 2008). The medication has been shown to be safe in children

and adolescents and helpful in reducing anxiety around meals as well as preoccupation with body image and weight in these age groups (Boachie, Goldfield, & Spettigue, 2003; Mehler et al., 2001). An NIH-funded study is currently underway to more conclusively determine the effectiveness of olanzapine in these measures as well as its side effect profile in patients with AN (National Institutes of Health, 2015).

Finally, smaller-scale studies have suggested the possible effectiveness of the mood stabilizer lithium (Gross et al., 1981), the opiate antagonist naltrexone (Marrazzi, Bacon, Kinzie et al., 1995), the antihistaminic and antiserotonergic agent cyproheptadine (Goldberg, Halmi, Eckert et al., 1979), and the mineral zinc (Birmingham, Goldner, & Bakan, 1994). Again, further investigation in the form of larger-scale studies is needed to investigate these medications.

Relapse Prevention

Weight restoration is only the first step in treatment of AN, and relapse prevention is essential for management of the illness. Treatment challenges and often mitigates patients' distortion of body image and excessive concern about shape and weight. It also interrupts rigid food rules and restriction of eating. However, because these symptoms are rarely eradicated, the emotional and behavioral features of AN continue to require attention after weight is restored.

Intensive programs consider prior treatment, whether effective or not, when planning for discharge and warn that vigilance monitoring eating behaviors and both purposeful and accidental weight loss is vital. They coach patients to effectively respond to a setback and use it as an opportunity to reach out for help rather than let it precipitate a relapse. Discharge plans often involve regular meetings between the patient and a clinical support team that may include a therapist, nutritionist, and/or psychiatrist.

More studies are needed to determine successful forms of relapse prevention, but evidence suggests that outpatient CBT is more effective than some commonly used strategies such as nutritional counseling (Pike, Walsh, Vitousek, Wilson, & Bauer, 2003). In addition, studies concerning predictors of recovery have found that patients who achieve a higher BMI during the acute phase of weight restoration, those who maintain their weight immediately after discharge from an intensive program, and those who continue to be motivated to recover during refeeding programs are the most successful in recovery. Further study is needed to determine if altering these factors, for example, by asking patients to get to a higher BMI during treatment, will lead to more successful relapse prevention (Kaplan et al., 2009; Carter et al., 2012).

Bulimia Nervosa

The treatment for BN has many similarities to that of AN. It aims to normalize eating behaviors and to redirect overemphasis of shape and weight. Treatment similarly begins with an assessment of symptom and weight history as well as current behaviors and comorbidities (Walsh, 2008).

BN appears to be more responsive than AN to psychological and pharmacologic treatments. Treatment for BN is usually delivered in outpatient settings, with higher levels of care being reserved for individuals who manifest electrolyte disturbances or other medical complications of illness, for those who have complicated psychiatric presentations including co-occurring conditions, and for those who have failed to respond to outpatient treatment.

Unlike AN patients, BN patients often report distress associated with their symptoms and may be more motivated to pursue treatment. Furthermore, these patients more commonly align with their providers around treatment goals. The major challenges associated with clinical management of BN include the secretive nature of the behaviors in some, especially younger patients, and the possibility of electrolyte disturbance or degree of medical compromise that could require multimodal treatment.

Psychotherapy

CBT treatment for BN is a structured psychotherapy with a large evidence base, generally offered over the course of three to 6 months (Fairburn,

Jones, Peveler, Hope, & O'Connor, 1993). The treatment includes approximately 20 individual sessions, held once or twice weekly, and aimed at normalizing eating behaviors by encouraging regular meals and snacks, redirecting the patient's rigid rules about eating, and reducing distorted thoughts about shape and weight (Wilson, Fairburn, Agras, Walsh, & Kraemer, 2002). About 25–50% of patients are able to abstain from binge and purge behaviors during their treatment, and many are able to maintain this state of recovery following treatment. Guided self-help manuals based on the principles of CBT have also been shown to be an effective treatment for BN (Wilson & Zandberg, 2012).

Family-Based Treatment

FBT has been adapted for the treatment of BN with a modest evidence base for adolescents. It has many similarities to FBT for AN: externalizing the illness and empowering the parents to restore their child to health (Le Grange, Crosby, Rathouz, & Leventhal, 2007; Le Grange & Lock, 2007). The first of three phases of FBT for BN typically consists of 2–3 months of weekly sessions aimed at interrupting binge/purge behaviors and restrictive dieting as well as any other pathological weight control behaviors through meal and post-meal supervision. The second phase, which occurs when the adolescent is no longer engaging in disordered behaviors, consists of biweekly therapy sessions in which the parents begin to return control of eating back to the patient. Finally, the third phase of treatment consists of monthly sessions that focus on assisting the family and the patient through the developmental process, specifically targeting areas that may have been affected by the illness.

Pharmacotherapy

A number of antidepressants have been shown to be helpful in curbing BN urges and behaviors, and fluoxetine is approved by the Food and Drug Administration (FDA) for therapeutic use for BN (Golden & Attia, 2011). It has a large evidence base in adults at 60 mg/day and a high tolerability rate (Shapiro et al., 2007). It has been shown to be beneficial in BN regardless of baseline mood status (Goldstein, Wilson, Ascroft, & Al-Banna, 1999), and a response is often demonstrated within 3 weeks (Sysko, Sha, Wang, Duan, & Walsh, 2010). Although a BN randomized controlled trial for fluoxetine has not been completed in children or adolescents, an open-label trial has shown that it is efficacious and well tolerated in these age groups, and the FDA has approved its use in these populations for depression (Kotler, Devlin, Davies, & Walsh, 2003; Golden & Attia, 2011).

Other medications have been shown to be effective in the adult population. These include tricyclic antidepressants (Mitchell & Groat, 1984; Hughes, Wells, Cunningham, & Ilstrup, 1986), monoamine oxidase inhibitors (Kennedy et al., 1993; Walsh, Stewart, Roose, Gladis, & Glassman, 1985), bupropion (Horne et al., 1988), the antiemetic medication ondansetron (Faris et al., 2000), and the anticonvulsant with appetite-suppressant and mood-stabilizing properties topiramate (Nickel et al., 2005). However, these medications have less of an evidence base than fluoxetine and/or are associated with side effects that limit their acceptability (Shapiro et al., 2007). Bupropion specifically is associated with grand mal seizures in BN patients (Horne et al., 1988).

Relapse Prevention

Treatment for BN is often successful at normalizing eating behaviors, interrupting rigid food rules, and terminating binge/purge behaviors. However, the emotional features of BN, including concern about shape and weight and history of entrenchment in disordered behaviors, leave patients vulnerable to relapse.

Indications of relapse have thus far proven to be severity of the illness and shorter length abstinence from behaviors following treatment (Halmi et al., 2002). Those with higher levels of preoccupations and rituals regarding their eating, higher levels of restraint regarding disordered thoughts and behaviors, and less motivation for change are at higher risk for relapse. Since these are predictors that can be measured, clinicians may be able to use this information to alter or increase the level of intervention and prevent lapses from precipitating

relapses. Alternative strategies, such as an intervention through mobile text messages, planned return visits, or phone calls, may be useful tools to maintain continued patient engagement after cessation of treatment (Robinson et al., 2006; Mitchell et al., 2004). Finally, continued use of fluoxetine in those who responded to acute treatment with the medication is useful in preventing relapse (Romano, Halmi, Sarkar, Koke, & Lee, 2002).

Prognosis and Outcome

Complete psychological and physical recovery rates are generally thought to be somewhere around 33% for those with AN and 74% for those with BN (Herzog et al., 1999). While it is impossible to predict an eating disorder patient's outcome with certainty, some patterns have been shown to be reliable indicators of prognosis.

Prognosis is better for those who are diagnosed and treated early, as indicated by the success of family-based treatment in AN (75–90% recovery) (Eisler et al., 1997; Walsh, 2008). Patients with AN who are able to remain at or above their goal weight for a period of 1 year are shown to do significantly better years after discharge than those who lose weight (even if only a small amount) immediately after treatment. Prepubertal onset, more severe symptoms, and significant comorbidities may be additional indicators of poorer outcomes in AN and BN patients.

Even among those who recover from AN or BN, some continue to live an impaired life after treatment, continuing to engage in eating-disordered behaviors and/or thought patterns (including preoccupations or rituals relating to food or body image, social isolation, and/or binging or purging behaviors). Those who are chronically ill may follow a pattern of remission followed by relapse over many years.

Rates of mortality in AN are higher than in any other psychiatric illness, with a crude mortality rate of about 5% per decade due to medical complications or suicide (for which rates are about 12 per 100,000 per year) (APA, 2013). The crude mortality rate for BN is lower, around 2% per decade (APA, 2013).

Conclusion

In summary, early recognition and treatment of AN and BN is crucial to positive prognosis. Clinicians who treat vulnerable populations especially must be informed about the symptom presentation in order to properly identify these illnesses, medically monitor their patients, and refer them to appropriate providers for specialized care. There is substantial evidence for effective treatment of both illnesses, especially in patients with a shorter duration of illness, namely, family-based treatment for children and adolescents with AN and fluoxetine and cognitive behavioral therapy for those with BN.

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Binge Eating Disorder

18

Erin E. Reilly, Lisa M. Anderson, Lauren Ehrlich, Sasha Gorrell, Drew A. Anderson, and Jennifer R. Shapiro

Recent estimates suggest that a large number of youth, particularly obese youth, will experience some form of binge eating at some point during childhood and adolescence (Tanofsky-Kraff, 2008). Though there still exists relatively little research on binge eating disorder (BED) in youth compared to other eating disorders (EDs), binge eating in youth has been prospectively linked to comorbid psychopathology, physiological concerns, and decreased quality of life (e.g., Tanofsky-Kraff et al., 2011). This chapter outlines the existing knowledge regarding BED and related behaviors in child and adolescent populations. We first provide an introduction to the disorder including relevant terminology, present the criteria for BED, and discuss relevant assessment tools for those working with youth that may binge eat. Next, we present a synthesis of existing research regarding the etiology of binge eating, as well as symptom presentation, course, diagnostic criteria, and treatments. We focus on recent advances in research regarding the presentation of binge eating and BED among youth and highlight gaps in existing research that indicate the direction of future work.

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An Introduction to BED

The Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association [APA], 2013) currently lists BED as a stand-alone ED diagnosis. Previously, those who reported problematic binge eating without any compensatory behaviors received a diagnosis of eating disorder, not otherwise specified (ED-NOS). Extensive literature has indicated that classifying BED as a distinct eating disorder category offers superior clinical utility for clinicians than information provided by an ED-NOS diagnosis (e.g., Hudson, Coit, Lalonde, & Pope, 2012; Striegel-Moore & Franko, 2008; Wilfley, Bishop, Wilson, & Agras, 2007; Wonderlich, Gordon, Mitchell, Crosby, & Engel, 2009). The specific DSM-5 criteria for BED are presented in Table 18.1. Notably, however, the current DSM-5 criteria may best capture binge eating psychopathology among adults, but do not list developmental considerations for youth. This chapter will present and discuss various proposed research criteria and developmental considerations for binge eating among younger populations.

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Table 18.1 Diagnostic and research criteria for binge eating disorder and loss of control eating disorder

Binge eating disorder (BED) DSM-5 diagnostic criteria[a]
A. Recurrent episodes of binge eating. An episode of binge eating is characterized by both of the following:
1. Eating, in a discrete period of time (e.g., within any 2-h period), an amount of food that is definitely larger than what most people would eat in a similar period of time under similar circumstances
2. A sense of lack of control over eating during the episode (e.g., a feeling that one cannot stop eating or control what or how much one is eating)
B. The binge eating episodes are associated with three (or more) of the following:
1. Eating much more rapidly than normal
2. Eating until feeling uncomfortably full
3. Eating large amounts of food when not feeling physically hungry
4. Eating alone because of feeling embarrassed by how much one is eating
5. Feeling disgusted with oneself, depressed, or very guilty afterward
C. Marked distress regarding binge eating is present
D. The binge eating occurs, on average, at least once a week for 3 months
E. The binge eating is not associated with the recurrent use of inappropriate compensatory behavior as in bulimia nervosa and does not occur exclusively during the course of bulimia nervosa or anorexia nervosa
Provisional BED research criteria for children[b]
A. Recurrent episodes of binge eating. An episode of binge eating is characterized by both of the following:
1. Food seeking in the absence of hunger (e.g., after a full meal)
2. A sense of lack of control over eating (e.g., endorse that, "When I start to eat, I just can't stop")
B. Binge episodes are associated with one or more of the following:
1. Food seeking in response to negative affect (e.g., sadness, boredom, restlessness)
2. Food seeking as a reward
3. Sneaking or hiding food
C. Symptoms persist over a period of 3 months
D. Eating is not associated with the regular use of inappropriate compensatory behaviors (e.g., purging, fasting, excessive exercise) and does not occur exclusively during the course of anorexia nervosa or bulimia nervosa
Loss of control eating disorder (LOC-ED) in children research criteria[c]
A. Recurrent episodes of LOC eating. An episode of LOC eating is characterized by both of the following:
1. A sense of lack of control over eating
2. Food seeking in the absence of hunger or after satiation
B. The LOC eating episodes are associated with three or more of the following:
1. Eating in response to negative affect
2. Secrecy regarding the episode
3. Feelings of numbness (lack of awareness) while eating
4. Eating more, or the perception of eating more, than others
5. Negative affect following eating (e.g., shame/guilt)
C. The LOC eating episodes occur, on average, at least twice a month for 3 months
D. Eating is not associated with the regular use of inappropriate compensatory behaviors and does not occur exclusively during the course of anorexia nervosa, bulimia nervosa, and binge eating disorder

Note: [a]APA (2013); [b]Marcus and Kalarchian (2003); [c]Tanofsky-Kraff et al. (2008)

Key Terms and Definitions

Those working with children and adolescents who meet sub- or full-threshold BED criteria should be aware of various distinctions between types of eating that are often considered during assessment. Often terms related to overeating or binge eating are used interchangeably and inconsistently. Thus, to maintain clarity throughout this chapter, we will define key terms:

Binge Eating Consuming an objectively large amount of food while feeling a loss of control (i.e., feeling that one cannot stop eating or is not in control of eating tempo, amount, or another characteristic that would lend itself to being uncontrollable). Binge eating is not limited to BED; binge eating is a behavior that has been observed among individuals diagnosed with AN, BN, and other specified feeding and eating disorders (OSFED).

Binge Eating Episode A single, discrete period of time during which an individual consumes a large amount of food while experiencing a feeling of loss of control. Binge eating episodes that meet both the amount and loss of control criteria have also been referred to as *objective* bulimic episodes (OBEs; Fairburn, 2008). In contrast, *subjective* bulimic episodes (SBEs; Fairburn, 2008) are characterized by feelings of loss of control while consuming a small to moderate amount of food over a discrete period of time.

Overeating Episodes (OEs) OEs refer to the act of consuming an objectively large amount of food in the absence of a loss of control (Fairburn, 2008).

Emotional Eating (EE) Eating in response to the experience of intense emotion (internally motivated eating). EE can occur in response to both positive and negative emotion (e.g., Geliebter & Aversa, 2003); however, studies of EE among youth have typically focused on negative emotion states (i.e., Tanofsky-Kraff, Theim, et al., 2007). Although not all EE leads to binge eating episodes (e.g., the amount of food consumed may not be objectively large or one may feel in control of eating), EE may lead to overeating behaviors (e.g., Nguyen-Michel, Unger, & Spruijt-Metz, 2007).

Eating in the Absence of Hunger (EAH) A form of behavioral disinhibition; eating when one is not physically hungry (e.g., food seeking after consuming a full meal). This behavior may indicate a tendency to make decisions to eat based primarily on external cues (i.e., cues other than internal or physiological hunger or satiety signals; Fisher & Birch, 2002).

Loss of Control Eating (LOC) LOC eating may occur when an individual begins to eat a snack or meal (in response to hunger cues) and continues to eat past the point of satiation. LOC episodes include all episodes during which the individual reports a loss of control regardless of whether the amount of food eaten is unambiguously large (Tanofsky-Kraff et al., 2011).

Diagnostic Criteria for Binge Eating Disorder Among Children and Adolescents

Prevalence Rates for BED Using Current DSM-5 Criteria

Some work has shown low prevalence rates of BED in childhood (i.e., 1–3.1% for full-threshold BED; Johnson, Rohan, & Kirk, 2002; Neumark-Sztainer, Story, Hannan, & Rex 2003). Other reports, however, indicate that binge eating behaviors commonly occur within this population (Swanson, Crow, Le Grange, Swendsen, & Merikangas, 2011). Prevalence rates for early-onset BED among youth appear to increase with pubertal development and age (Field et al., 1999).

Limitations Related to the Use of the Current DSM-5 Criteria

Youth diagnosed with early-onset BED (under 18 years old) have reported prior onset of subclinical binge eating behaviors at an average age of 12.8 years (Marcus, Moulton, & Greeno, 1995). Thus, subthreshold levels of binge eating may predict later fulfillment of BED criteria. While OBEs are reported by youth (e.g., Swanson et al., 2011), these episodes typically occur infrequently. The infrequent incidence of OBEs among youth points to two possibilities: one possibility is that BED is uncommon in youth. However, it is also possible that the presentation of BED may differ in youth and using criteria developed from adult samples to diagnose children and adolescents may result in under-identification of clinically relevant binge eating behaviors.

Although LOC eating does not fulfill criteria for BED, LOC is linked to overeating and impaired eating control, increased anxiety, depression, and body image dissatisfaction (e.g., Goossens, Braet, & Decaluwé, 2007; Hilbert & Czaja, 2009). Thus, LOC eating in itself may be a salient indicator of clinical levels of psychological distress among youth.

Other Criteria Sets for Diagnosing Child and Adolescent Binge Eating Disorder

Although researchers have acknowledged that children and adolescents can be diagnosed with full-threshold BED (Johnson et al., 2002; Morgan et al., 2002), it has been suggested that broader diagnostic criteria for youth should be considered as compared to the current DSM-5 criteria (e.g., Bravender et al., 2010; Marcus & Kalarchian, 2003; Tanofsky-Kraff, Marcus, Yanovski, & Yanofski, 2008). Several research teams have recommended provisional criteria sets for diagnosing BED and loss of control eating disorder (LOC-ED) among youth (e.g., Bryant-Waugh & Lask, 1995; Marcus & Kalarchian, 2003; Tanofsky-Kraff et al., 2008).

Proposed Research Criteria for BED and LOC-ED Among Youth Bravender et al. (2007) presented the Workgroup for Classification of Eating Disorders in Children and Adolescents (WCEDCA), which suggested methods for assessing and diagnosing BED among youth that account for developmental stage (i.e., level of insight or veracity of self-report). The WCEDCA suggested that diagnostic criteria for BED among youth should employ lower and more developmentally sensitive symptom thresholds for binge eating behaviors (e.g., at least one episode of LOC eating within the past 3 months), rather than the more stringent OBE criteria used for adults (see Table 18.1 for criteria in DSM-5, as well as proposed research criteria for BED and/or LOC-ED among youth).

Marcus and Kalarchian's (2003) proposed research criteria – developed specifically for identifying problematic binge eating among youth – did not include a criterion requiring that a child or adolescent report regular consumption of an objectively large amount of food. The frequency specified within their proposed criterion allowed for fewer binge eating behaviors, thus allowing for more flexibility with symptom presentation (see Marcus & Kalarchian, 2003 for provisional BED research criteria proposal). Most recently, Tanofsky-Kraff et al. (2008) proposed research criteria for a "loss of control eating disorder" (LOC-ED) for children (12 or younger; see Table 18.1). The authors acknowledged that their frequency and duration criteria were arbitrarily set, due to a general lack of research to support and guide the suggested criteria. Thus, research is needed to determine the validity and clinical utility of the suggested criteria.

Assessment Tools and Methods for Children and Adolescent BED

General Considerations There are a number of reliable methods for assessing the behavioral components of BED among adults, including self-report measures and direct observation (Tanofsky-Kraff et al., 2013). However, there are fewer methods for reliably evaluating BED among youth (Brownley, Shapiro, Bulik, 2009; Bulik, Brownley, & Shapiro, 2007). Assessment methods developed and used for adult samples may not be reliable or applicable for younger individuals, and differences may exist in identification of binge eating across different measurement types. In particular, children and adolescents may struggle to identify binge eating episodes when completing self-report measures (Decaluwé & Braet, 2004). These authors hypothesized that the problems experienced by the children were partially due to the inability to ask for more detailed information regarding the identification of binge eating episodes. Differences between assessment methods were significant, such that the self-report measure failed to identify three cases of binge eating among a sample of 139 youth seeking inpatient treatment for obesity, while also identifying 40 children as binge eaters for whom the interview had not. Thus, when conducting assessments among youth, researchers and clinicians should consider assessment modality. Interview-based assessments may be most useful in identifying binge eating behaviors among youth (Birgegård, Norring, & Clinton, 2014).

Structured Interviews Noting the lack of assessments for BED in children, Shapiro et al. (2007) developed the Children's Binge Eating Disorder Scale (C-BEDS) – a structured interview assessment

designed to be developmentally sensitive, both in its content and administration. Shapiro and colleagues compared the C-BEDS to the Structured Clinical Interview for the Diagnosis of DSM-IV Disorders BED module (SCID). Noted in their validation study, the majority of their sample had difficulty recollecting the age of onset for specific behaviors. Thus, the C-BEDS assessment criteria were amended to omit the age of onset. Shapiro et al. (2007) found that approximately half of the children reported eating in the absence of hunger, LOC eating, and desiring food as a reward for accomplishing a task or performing well. Additionally, almost two-thirds of the children reported eating in response to negative emotions – a behavior associated with BED among adults. Overall, the authors concluded that the C-BEDS captured a larger range of children who endorsed problematic binge eating as compared to the SCID. After broadening the SCID criteria to include sub-syndromal individuals, the C-BEDS identified similar numbers of child BED cases. Thus, while the number of identified cases did not differ, the authors suggested that the C-BEDS appeared to offer a child-friendly, understandable assessment tool that was developmentally sensitive to binge eating behaviors and related pathology among youth.

Self-Report Measurements The ChEDE, based off the widely used Eating Disorder Examination Questionnaire (EDE-Q; Fairburn & Cooper, 1993) for children, is a self-report measure that evidences good psychometric properties (Goldschmidt et al., 2007). In addition, Tanofsky-Kraff et al. (2008) developed a self-report questionnaire, Eating in the Absence of Hunger for children and adolescents (EAH-C). Because the EAH construct is generally considered to be similar to LOC eating, this assessment provides clinicians and researchers with a useful self-report tool to assess EAH among youth. Altogether, several child-specific assessments have been designed that allow for better assessment of binge-eating-related constructs (i.e., EAH) in more developmentally sensitive modalities (i.e., interview assessments versus self-report questionnaires). When possible, clinicians should use clinical interviews and developmentally sensitive measures to assess for BED among youth. This will ensure better identification of the subgroup of children who report binge-eating-related pathology, such as clinically meaningful LOC eating or distress related to subthreshold frequencies of OBEs.

Etiology

Biological Factors

Research on the biological underpinnings of BED has predominantly been conducted using adult samples and animal models (e.g., Mathes, Brownley, Mo, & Bulik, 2009). Nonetheless, these findings, as well as the few examinations of younger samples, yield insight regarding potential genetic or neurological factors underlying the development of LOC and binge eating in children and adolescents.

Genetics Heritability estimates from population-based twin studies and adoption work suggest that a latent vulnerability to binge eating may fall anywhere between 41 and 82% (Bulik, Sullivan, & Kendler, 1998; Bulik, Sullivan, & Kendler, 2003; Helder & Collier, 2011; Klump, Suisman, Burt, McGue & Iacono, 2009). Candidate genes and related polymorphisms posited to be particularly relevant to binge eating symptomatology consistently relate to the serotonergic and dopaminergic neurotransmitter systems (e.g., Scherag, Hebebrand, & Hinney, 2010). Specifically, mutations involving the melanocortin-4 receptor gene (MC4R; Tao & Segaloff, 2003; Valette et al., 2012) and the fat mass and obesity-associated gene rs9939609 (Tanofsky-Kraff et al., 2009) have been linked to obesity and LOC eating in children and adolescents. In studies of adults, genes associated with the expression of brain-derived neurotrophic factor (BDNF) have also been associated with binge eating (Monteleone, Tortorella, Castaldo, & Maj, 2006; Rosas-Vargas, Martinez-Ezquerro, & Bienvenu, 2011).

A comprehensive understanding of the genetic underpinnings of binge eating will likely include an emphasis on epigenetics (Campbell, Mill, Uher, & Schmidt, 2011). In a recent review on

gene x environment interactions in the etiology of disordered eating and obesity, Campbell et al. (2011) reviewed findings that environmental factors such as early life stress (e.g., Danese, Pariante, Caspi, Taylor, & Poulton, 2007) and maternal nutrition (e.g., Reynolds, Godfrey, Barker, Osmond, & Phillips, 2007) contribute to epigenetic changes that influence risk for disordered eating. Campbell et al. (2011) posited that some of the previously mentioned genes for LOC and binge eating (e.g., FTO) are subject to epigenetic processes. There is also evidence of epigenetic changes in dopaminergic genes in individuals with EDs (Frieling et al., 2010). It is important to note that these findings are in their infancy; in addition, most work examining epigenetic risk for obesity and disordered eating has not specifically targeted risk for binge eating. Therefore, as methods in epigenetics move forward, it will be important to determine whether specific gene x environment interactions best predict risk for binge eating.

Hormones and Appetitive Response Work examining the role of appetitive hormones such as insulin, leptin, ghrelin, glucagon, and cholecystokinin in binge eating has identified several general irregularities among samples of both children and adults (Geliebter, Yahav, Gluck, & Hashim, 2004). However, these irregularities may be attributable to an individual's overweight status, rather than binge eating per se (Adami et al., 1995; Lourenço et al., 2008). For example, larger gastric capacity associated with BED may influence the irregular release of appetitive hormones observed in these individuals (e.g., Geliebter et al., 2004). Research in adult samples has shown higher postprandial insulin among individuals who binge eat (Geliebter et al., 2004; Russell, Hooper, & Hunt, 1996), and dysregulated levels of leptin and ghrelin among obese individuals as well as those who binge eat (Campfield, 2002; Geliebter et al., 2004; Tschöp et al., 2001), though it remains unclear how these differences might emerge. Other work has found no such differences; for instance, one investigation of adolescents found no differences in appetite-regulating hormones between those individuals who binge ate and those who did not (Adamo et al., 2014).

One potential explanation for discrepant findings regarding biological differences in satiety and hunger may be that there exist distinct subgroups of overweight and obese individuals with varying hormonal processes in response to food consumption or hunger. For example, using self-report measures and behavioral paradigms, Boutelle and colleagues (2014) identified three distinct subgroups of overweight and obese children with different levels of satiety response. Further investigation into the nuances of appetite response is needed to clarify their relation to LOC/binge eating among youth.

Neurocognition The majority of research on neurological functioning relevant to BED has examined differences between obese and non-overweight individuals rather than differences between those who report and do not report binge eating. Current research highlights pathways involved in inhibitory processes (e.g., the frontal cortex), emotions (e.g., the amygdala), and the reward value of food (e.g., the striatum and orbitofrontal cortex) as key areas in mediating responses to food cues (Holsen et al., 2005; Killgore & Yurgelun-Todd, 2006; Michaelides, Thanos, Volkow, & Wang, 2012; Nederkoorn, Braet, Van Eijs, Tanghe, & Jansen, 2006; Schienle, Schäfer, Hermann, & Vaitl, 2009; Stice et al., 2011; Wang, Volkow, Thanos, & Fowler, 2004). Most notably, compared to non-overweight adolescents, obese youth demonstrate hyperactivity of the reward circuits in response to food cues and do not modulate response of the reward and satiety circuits following a meal (Bruce et al., 2010; Stice, Davis, Miller, & Marti, 2008; Stice et al., 2011). Stice and colleagues (2011) found that non-overweight youth at high risk for obesity show increased striatal activation prior to food intake and response to food-related cues, suggesting that those at risk for obesity show hypersensitivity to food-related reward, which may increase the chance of overeating. Chronic activation of this area over time could conceivably lead to downregulation of the reward circuit and the dysregulated striatal response observed in obese youth. In a study comparing differing neural responses to sucrose and water between obese and healthy-weight children, Boutelle and colleagues

(2014) found no differences in striatal activation. However, they did find that obese children showed increased activation in emotional areas (i.e., amygdala, hippocampus) in response to glucose as compared with the healthy-weight controls.

A recent study found that obese adolescents reported higher disinhibition, lower performance on cognitive tests gauging executive function and self-regulation, and volume reductions in the frontal lobe, as compared to lean adolescent controls (Duchesne et al., 2010; Maayan, Hoogendoorn, Sweat, & Convit, 2011). Lower scores on these cognitive tests indicate that obese teenagers may experience difficulties with impulsivity, planning, and inhibiting primary responses in favor of responses more oriented to long-term goals; any or all of these difficulties could play some role in a propensity to overeat or binge eat.

In sum, research suggests that youth who are obese (and perhaps most at risk for overeating and LOC eating) evidence distinct neurocognitive profiles in areas related to self-regulatory, emotional-, and satiety-based behaviors. More research is needed to determine if these differences are more salient to a child or adolescent's overweight status or relate specifically to LOC and binge eating behaviors.

Emotional and Psychological Factors

The majority of research examining emotional and psychological risk for BED has been conducted in adults, with few studies conducted with youth. Although several of the risk factors for LOC and binge eating (e.g., depression) are also associated in cross-sectional work with BED, the following section will focus predominantly on the relevant variables identified through longitudinal work. Longitudinal assessment provides the clearest picture of variables relevant to the etiology, rather than consequences, of LOC and binge eating.

Depression and Negative Affect Research suggests that depressive symptoms may serve as a risk factor for the development of LOC and binge eating, as negative affect consistently promotes and precedes binge eating (Greeno, Wing, & Shiffman, 2000; Haedt-Matt & Keel, 2011; Stice & Agras, 1998). Additionally, negative affect is featured in some of the most notable etiological models of disordered eating (e.g., Hawkins & Clement, 1984; Heatherton & Baumeister, 1991; Stice, 2001). Within these models, binge eating and other disordered eating behaviors serve as a means to regulate negative affect.

The relation between negative affect and binge eating has been supported by prospective research; a large, longitudinal cohort study found that females who reported depressive symptoms at baseline were twice as likely as their peers to report beginning to overeat at 4-year follow-up (Skinner, Haines, Austin, & Field, 2012). Also, Stice, Presnell, and Spangler (2002) found that depression was prospectively linked to binge eating in adolescent girls, including those who reported low levels of appearance overvaluation (e.g., Johnson, Cohen, Kasen & Brook, 2002; Stice, Killen, Hayward, & Taylor, 1998).

Emotion Regulation Research has demonstrated that both youth and adults with LOC or binge eating show difficulties with emotional regulation (Czaja, Rief, & Hilbert, 2009; Whiteside et al., 2007). Studies have found that individuals who report negative affect and maladaptive emotion regulation are more likely to overeat or report emotional eating (Evers, Stok, de Ridder, 2010; Spoor, Bekker, Van Strien, & van Heck, 2007). Additionally, eating in order to cope with negative emotions prospectively predicts the onset of binge eating (Allen, Byrne, La Puma, McLean, & Davis, 2008). In contrast, a recent study using ecological momentary assessment found no evidence that LOC eating serves an affect-regulation function in children (Hilbert, Rief, Tuschen-Caffier, de Zwaan, & Czaja, 2009). Therefore, emotion regulation may play a different role in binge eating depending on an individual's developmental stage.

Impulsivity Research consistently shows that youth who are obese and report binge eating show higher levels of impulsivity than do healthy-weight controls (Nederkoorn et al., 2006). Children who report LOC eating also report lower levels of self-directedness and cooperation, in combination with higher levels of impulsivity (Hartmann, Czaja, Rief, & Hilbert, 2010). A recent longitudinal study of elementary school children

found that negative urgency (a facet of impulsivity, reflecting a tendency to act rashly when distressed) was associated with the development of an expectancy that eating would alleviate negative mood states. These affect-driven eating expectancies subsequently predicted later binge eating behavior (Pearson, Combs, Zapolski, & Smith, 2012). This suggests that impulsivity may be most relevant to binge and LOC eating when considered alongside negative affect and eating expectancies.

Self-Esteem Low self-esteem has been associated with increased likelihood of binge eating over time in females (Stice et al., 2002), and similarly elevated in individuals with BED of both genders (Tanofsky, Wilfley, Spurell, Welch, & Brownell, 1997). Additionally, research has found that self-esteem may interact with other risk factors, such as dietary restraint, to predict the disinhibited state often observed in binge eating (Polivy, Heatherton, & Herman, 1988). One study of adult females with BED has shown that low self-esteem seems to mediate the relation between self-criticism and overvaluation of shape and weight (Dunkley & Grilo, 2007). However, such investigations have not yet been completed in samples of youth.

Dietary Restraint and Dieting Etiological models of disordered eating (e.g., the Dual Pathway Model, Stice, 2001; Restraint Theory, Polivy & Herman 1985; Transdiagnostic Model of ED; Fairburn, 2008) suggest that dieting (i.e., engaging in active restriction of food or food groups) and dietary restraint (i.e., the cognitive efforts to limit caloric intake) mediate the relation between body dissatisfaction and disordered eating behaviors. Although some research suggests that relations between dieting and binge eating exist in adolescents (Dakanalis et al., 2014; Stice, 2001; Stice & Agras, 1998; Stice, Shaw, & Nemeroff, 1998) and children (Allen, Byrne, Oddy, & Crosby, 2013), others report no evidence for the link (Spoor et al., 2006; Stice, 1998; Stice, Martinez, Presnell, & Groesz, 2006).

These discrepancies within the literature may reflect the presence of different subgroups of children who report LOC eating (Tanofsky-Kraff, Faden, Yanovski, Wilfley, & Yanovski, 2005). That is, there may exist several trajectories through which dieting and binge eating develop and are linked. For example, individuals that report dieting before the onset of binge eating fall most closely in line with Restraint Theory's temporal relations between dieting, disinhibition, and binge eating (Polivy & Herman, 1985). However, within a sample of treatment-seeking overweight adolescents, binge eating prior to dieting was more common than dieting prior to binge eating (Tanofsky-Kraff et al., 2005). Other work has also found that binge eating prior to dieting is associated with higher lifetime psychopathology than dieting prior to binge eating (Spurrell, Wilfley, Tanofsky, & Brownell, 1997).

History of Disordered Eating In addition to or other than initial dietary restraint, children at risk for later binge eating may have a history of engaging in other disordered eating behaviors. These include EAH (Fisher & Birch, 2002), secretive eating or hiding food (Stice, Agras, & Hammer, 1999), eating to manage negative affect (Stice et al., 2002), and overeating with no loss of control (Stice et al., 1999). The exact mechanism through which various eating behaviors lead to full-scale BED remains unclear, however.

Social Factors The social context seems to play a role in the etiology of binge eating among children and adolescents. Additionally, it is likely that these variables interact with other factors (e.g., biological, emotional) to exacerbate the risk for LOC and overeating.

Family Factors Several variables related to the family have been posited as relevant to the development of LOC and binge eating. Similar to other disordered eating behaviors, a child is more likely to report LOC eating if their mother (Zocca et al., 2011) or father (Goldschmidt et al., 2014) also report binge eating behaviors. The mealtime rituals and feeding practices of a family appear to be important in the etiology of problematic eating patterns in youth. For instance, a recent investigation of a treatment-seeking sample of

children found that emotional feeding behavior by parents (i.e., the tendency to provide the child food following negative emotion) was the most salient predictor of emotional eating in children (Braden et al., 2014). Additionally, stringent parental restriction of food intake is linked cross-sectionally to binge eating in children (Carper, Fisher, & Birch, 2000; Fisher & Birch, 2002). Finally, parental criticism surrounding weight (Hartmann, Czaja, Rief, & Hilbert, 2012) and being perceived as overweight during middle childhood by one's parents, regardless of actual weight status, is longitudinally related to binge eating in late adolescence and early adulthood (Allen et al., 2013).

Other general family factors also appear relevant to the etiology of LOC and binge eating. For instance, children's attachment styles have been highlighted as an important factor accounting for the link between LOC eating and self-esteem, particularly in younger samples (Goossens, Braet, Bosmans, & Decaluwé, 2011). Insecure attachment relates to higher levels of LOC eating in a sample of children 8–11 years old; additionally, it seems that attachment mediates the relation between self-esteem and LOC eating (Goossens et al., 2011). Exposure to parental under-involvement, as well as familial arguments and depression, is associated with greater levels of childhood LOC eating (Hartmann et al., 2012). Lower levels of family connectedness have also been linked with binge eating in adolescent females (French et al., 1997).

Peer Factors Cross-sectional research on the relation between individual reports of binge or LOC eating and those of their peers has been mixed. Some studies have found that peers' binge eating predicts individuals' binge eating (Crandall, 1988; Eisenberg et al., 2012; Goldschmidt et al., 2014; Hutchinson & Rapee, 2007; Pike, 1995), while others have not (Meyer & Waller, 2001; Paxton, Schutz, Wertheim, & Muir, 1999). Greater levels of peer acceptance concerns are linked to binge eating (French et al., 1997). There have been no longitudinal studies examining peer influences on binge or LOC eating to date, limiting the conclusions that can be drawn from existing literature.

Interpersonal Difficulties The interpersonal model of BED (Wilfley, MacKenzie, Welch, Ayres, & Weissman, 2007) proposes that social problems result in negative affect and low self-esteem, which in turn serve as triggers for binge eating. Though the model was originally developed using adult samples, the interpersonal model was also validated in a sample of youth who reported LOC eating (Elliott et al., 2010). More generally, lack of perceived social support and interpersonal issues has also been prospectively linked with binge eating onset (Berger et al., 2014; Stice et al., 2002; Van Strien, Engles, Van Leeuwe, & Snoek, 2005).

Sociocultural Pressure and the "Thin Ideal" Sociocultural pressures to be thin have been identified as important influences in the etiology of eating disorders (Hausenblas et al. 2013). Specific to binge and LOC eating, body dissatisfaction that arises due to internalization of the thin ideal has been shown to predict binge eating in adolescent females (Stice, Ng, & Shaw, 2010; Stice & Shaw, 2002).

Symptom Presentation

Relevant Demographic Characteristics Across samples of youth, some studies have found that binge eating is more common in females (Croll, Neumark-Sztainer, Story, & Ireland, 2002; Goossens, Soenens, & Braet, 2009). Additionally, some work has suggested that regardless of overall prevalence, the binge eating reported by girls displays more maladaptive characteristics (Marcus & Kalarchian, 2003), such as greater embarrassment about binges (Whitaker et al., 1989) and increased loss of control (Sonneville et al., 2013; Tanofsky-Kraff et al., 2011).

Other work has suggested that binge eating is more common in males (Childress, Brewerton, Hodges, & Jarrell, 1993; Whitaker et al., 1989). While males generally report less eating disorder symptomatology as compared with females, a recent study of boys aged 12–18 years revealed significant evidence of reported LOC, as well as objective binge episodes. Of the 531 boys included in this sample, 6% reported regular episodes of

objective binge eating, and 8.3% reported regular episodes of loss of control eating. This study also found that loss of control eating was nearly twice as common in older adolescent boys than in younger adolescent boys (Mond et al., 2014). Thus, research outcomes are mixed; the prevalence of LOC and binge eating across genders in youth remains unclear.

Findings regarding the role of ethnicity and race in binge eating behaviors are also mixed. One study found that the behavior is most common in Hispanic females as compared to other ethnic groups (Croll et al., 2002), while another study suggested that binge eating is more common among African-American males as compared to White males and females (Johnson et al., 2002), and yet another study found no notable differences across ethnic/racial groups in binge eating (French et al., 1997).

Associated Impairments and Conditions

Several associated conditions and impairments linked to binge and LOC eating may be relevant to the development of maladaptive ED behaviors and treatment outcome. In youth, such impairments include physiological conditions (i.e., overweight, obesity, and metabolic syndrome), comorbid psychopathology, and interpersonal issues that may need to be explicitly addressed within the context of treatment.

Overweight and Obesity LOC eating is positively related to BMI and body fat mass (Tanofsky-Kraff, 2008), and reported binge and LOC eating longitudinally predict excessive weight gain in children and adolescents (Field et al., 2003; Stice, Cameron, Killen, Hayward, & Taylor, 1999; Tanofsky-Kraff et al., 2006; Tanofsky-Kraff et al., 2009). Overweight youth are more likely to report LOC eating than their peers (e.g., Allen et al., 2008). Not all overweight and obese children and teens report binge or LOC eating, however, and not all children who binge eat are overweight. Some studies have suggested that children seeking treatment for overweight and obesity who also report overeating episodes with LOC show higher BMIs than overweight children without LOC episodes (Decaluwé & Braet, 2003; Morgan et al., 2002). Other work has found no difference in BMI between overweight or obese children with LOC eating and those without (e.g., Lourenço et al., 2008; Mirch et al., 2006). It is possible that obese youth who endorse LOC eating represent a severe subtype of individuals who are younger, endorse more comorbid psychopathology, and show poorer progress in weight-loss treatment (Glasofer et al., 2007; Goossens et al., 2007; Isnard et al., 2003; Wildes et al., 2010).

Metabolic Syndrome (MetS) MetS, a constellation of metabolism-related factors that promote atherosclerotic cardiovascular disease, has been observed among adult obese populations (Ford, Giles, & Mokdad, 2004). It is only in the past several years that researchers have studied the prevalence of MetS in children. Tanofsky-Kraff et al. (2012) found that among children aged 5–12 years at high risk for adult obesity, binge eating was associated with 5.33 greater odds of having MetS at 14-year follow-up, as well as increased levels of triglycerides and visceral adipose tissue (i.e., fat tissue). Given that MetS is associated with higher levels of coronary heart disease and stroke (Isomaa et al., 2001), future research should continue to investigate the unique contributions of binge eating to the development of MetS, independent of weight status.

Emotion Regulation Though emotional regulation seems to play a role in the etiology of binge eating, it also represents an issue that may complicate an individual's clinical presentation and treatment. Using eating as an emotion regulation strategy may be specifically linked to overeating (Van Strien et al., 2005) and EAH (Moens & Braet, 2007) in children and adolescents.

Comorbid Psychopathology

Depression, Anxiety, and Suicide Attempts Though depression and negative affect are linked prospectively to binge eating, these symptoms may also

emerge in response to LOC or binge eating in children and adolescents. Children who report higher levels of broadly defined binge eating also endorse higher levels of negative mood, depressive symptoms, and characteristics of anxiety (Ackard, Newmark-Sztainer, Story, & Perry, 2003; Eddy et al., 2007; Glasofer et al., 2007; Ledoux, Choquet, & Manfredi, 1993; Morgan et al., 2002; Tanofsky-Kraff, 2008; Wildes et al., 2010). Prospective research has also shown that persistent LOC eating and overeating are linked with the emergence of depressive symptoms over time (Skinner et al., 2012; Tanofsky-Kraff et al., 2011). Although some work has found that depressive symptoms may be higher in children and adolescents meeting the threshold for full criteria BED compared to those reporting subthreshold or no binge eating (Glasofer et al., 2007; Johnson, Grieve, Adams, & Sandy, 1999), other work has found that *both* subthreshold and full-threshold binge eating are associated with similar levels of depressed mood (Ackard et al., 2003; Berkowitz, Stunkard, & Stallings, 1993; Isnard et al., 2003). Further, one study showed that over 25% of girls and boys who met criteria for binge eating syndrome had attempted suicide (Ackard et al., 2003). Thus, risk for internalizing problems and suicidal ideation may be elevated in those who report LOC and binge eating.

Externalizing Problems Particularly in adolescents, binge eating has shown to be cross-sectionally related to substance use and abuse (Croll et al., 2002; Ross & Ivis, 1999), as well as longitudinally related to the onset of binge eating (Sonneville et al., 2013). Across samples of children and adolescents, LOC eating is also linked to parent-reported behavioral problems (Glasofer et al., 2007; Tanofsky-Kraff et al., 2005), and externalizing problems seem to prospectively predict binge eating (Allen et al., 2013).

Low Self-Esteem Low self-esteem and self-image problems are common in youth who experience binge eating (Ackard et al., 2003; Decaluwé, Braet, & Fairburn, 2003; Goossens et al., 2009; Tanofsky-Kraff et al., 2005; Wildes et al., 2010). Some work has linked the low self-esteem observed in young children with LOC eating to other variables; for instance, Goossens et al. (2011) found that the relation between low self-esteem and LOC eating in preadolescence was explained by insecure attachment with parental figures.

Eating Disorder Psychopathology The "core psychopathology" of eating disorders, which includes overvaluation of shape and weight, eating concerns, and drive for thinness, is readily linked to episodes of binge eating within models well validated in adult samples (see the Transdiagnostic Theory of ED; Fairburn, Shafran, & Cooper, 2003). As such, recent work has begun to examine the links between these cognitive symptoms of EDs and binge eating in adolescents and children (e.g., Fitzsimmons-Craft et al., 2014). Although one study found that overvaluation of shape and weight did not prospectively predict the onset of binge eating (Allen et al., 2008), other research in adolescent samples has shown cross-sectional links between overvaluation of shape and weight and eating disorder symptoms, including binge eating (e.g., Ledoux et al., 1993; Tanofsky-Kraff et al., 2005; Wildes et al., 2010), LOC eating, and increases in disordered eating attitudes over time (Tanofsky-Kraff et al., 2011). This discrepancy in findings has been examined by recent studies. Some argue that the abstract reasoning involved in cognitive elements of disordered eating cannot occur within childhood samples (Bravender et al., 2007), while others argue that there may be meaningful subtypes of children with LOC eating who show different levels of overvaluation of shape and weight (Goldschmidt et al., 2011; Hilbert & Czaja, 2009).

Course and Developmental Challenges

Course of BED

LOC eating appears to be a relatively common experience in preadolescence, occurring in 9.3% of non-overweight and overweight, nontreatment-seeking children aged 6–12 (Tanofsky-Kraff et al., 2004). As mentioned previously, maladaptive

eating behaviors like LOC eating and OOE may begin much earlier than typical age of onset for BED and are central to the psychological disturbances that lead to the disorder (Colles, Dixon, & O'Brien, 2008; Tanofsky-Kraff et al., 2004). LOC eating in children often persists into adolescence and young adulthood (Tanofsky-Kraff et al., 2011) and tends to worsen over time (Sonneville et al., 2013).

Emerging evidence suggests that persistent LOC eating in children may significantly predict partial/full-syndrome BED at short-term follow-up (Hilbert & Brauhardt, 2014). Overeating in childhood may be relatively transient and not necessarily lead to further eating pathology, but it may be the *persistence* of LOC eating over time that becomes problematic (Allen et al., 2008). A study of children aged 8–13 years showed that stability of LOC over time predicted onset of BED over and above other risk factors, such as comorbid psychopathology or BMI (Hilbert & Brauhardt, 2014). In a population-based sample of 14-year-olds meeting criteria for a binge eating or purging disorder, nearly half (44%) failed to meet the criterion for an eating disorder at ages 17 and 20 (Allen et al., 2013). However, 45% of participants' meeting criteria for BED at age 14 developed compensatory behaviors and met full or partial criteria for BN by age 17.

A recent large-scale investigation of women revealed that childhood-onset binge eating (versus adult-onset binge eating) may lead to higher frequency of ED diagnoses (Brewerton, Rance, Dansky, O'Neil & Kilpatrick, 2014). Individuals who report earlier onset of binge eating behaviors also tend to meet criteria for BED at earlier ages, report more severe psychiatric disturbances than those reporting a later age of onset, and generally have poorer treatment outcome (Agras et al., 1995; Marcus & Kalarchian, 2003; Safer, Lively, Telch, & Agras, 2002). A longitudinal study of adolescents transitioning into young adulthood demonstrated that improvements in body satisfaction, decreases in depressive symptoms, and increases in self-esteem contributed to the cessation of binge eating over time (Goldschmidt, Wall, Loth, Bucchianeri & Neumark-Sztainer, 2014).

Children and adolescents who struggle with BED or subthreshold BED at any point in their lives are at increased risk for relapse following treatment compared to other EDs. One study found relapse rates for individuals with subthreshold/full BED to be 33%; this was slightly better than for bulimia nervosa (41%) but worse than for purging disorder (5%) (Stice, Marti, Shaw & Jaconis, 2009). Furthermore, adolescents with BED are highly unlikely to seek treatment for the disorder (Swanson et al., 2011).

Obesity and Related Developmental Challenges

As mentioned previously, there is limited research conducted with individuals who have BED and are healthy weight, suggesting that obesity is often associated with the course of the disorder. The majority of current research supports the finding that children with maladaptive eating behaviors such as LOC and OO are more likely to be obese (e.g., Allen et al., 2008; Tanofsky-Kraff et al., 2013).

Health Factors that Affect Development Health problems associated with binge eating pose particular challenges for children and adolescents. Sleep disturbance is commonly reported in overweight and obese individuals, and one study found greater disturbances in sleep-wake cycles among obese youth who reported engaging in binge episodes compared to obese youth who did not binge (Tzischinsky & Latzer, 2006). BED in children also precipitates the premature development of visceral adipose fat and worsening triglycerides; one study found that children 5–12 years old who self-reported binge eating were 5.3 times more likely to develop metabolic syndrome at 5-year follow-up (Tanofsky-Kraff et al., 2012).

Quality of Life In addition to health problems for individuals with BED that interfere with appropriate stages in development, binge eating also has a negative impact on quality of life. Not unique to BED, but across all eating disorder diagnoses, individuals who experience persistent ED symptoms in the ages of 14–20 are less likely to complete high school (Allen et al., 2013). One study of

children aged 12–17 found that when controlling for BMI, binge eating had a general negative impact of health-related quality of life (Pasold, McCracken & Ward-Begnoche, 2013). In this study, significant correlations between measures of fatigue and binge eating suggest that emotional and cognitive deficits associated with binge eating may be related to lower quality of sleep. Another study explored quality of life in obese adolescents via interview and questionnaire and found that, regardless of weight, binge eating significantly impaired quality of life in the domains of mobility, activities of daily living, work/school, social functioning, and self-esteem (Ranzenhofer et al., 2012).

Quality of life appears to be differentially associated with binge eating across demographic groups. Pasold et al. (2013) found that for African-Americans, binge eating was only related to the social aspects of health-related quality of life, while for Caucasians, all aspects of health-related quality of life (i.e., physical, psychological, social, school, emotional, fatigue) were significantly negatively related to binge eating. In the same study, binge eating had a greater negative impact on quality of life for females as compared to males and for individuals 14 years and older (versus younger than 13).

Treatment and Outcome

To date, there have been no clinical trials evaluating efficacy of treatments for clinical levels of BED in youth (Kelly, Shank, Bakalar, & Tanofsky-Kraff, 2014). The majority of studies related to binge eating evaluate interventions of binge and LOC eating in youth or BED in adults, rather than a clinical diagnosis of BED in youth (Wilfley, Vannucci, & White, 2010). The "gold standards" in treatment of adult BED [cognitive behavior therapy (CBT) and interpersonal therapy (IPT)] have been modified for youth in the treatment of overweight and LOC eating with some success (Wilson & Shafran, 2005); however, more work is needed to evaluate their efficacy and effectiveness in treating binge eating in youth.

Treatment of BED can target the patient's eating behavior, psychological well-being, or both. Maladaptive eating patterns may be targeted through behavioral weight-loss methods, such as diet and exercise changes. CBT also emphasizes regulated eating, self-monitoring, and effective problem-solving skills. However, this treatment should be focused on health, learning hunger, and satiety cues and decreasing binge eating rather than weight loss in order to prevent any potential consequential restrictive eating disorders that may arise from dieting. Other treatments aim to address the social support deficits and negative emotionality that may create vulnerability to binge eating (Tanofsky-Kraff et al., 2010).

Some evidence suggests that treatments that primarily address eating behaviors may result in more rapid and sustained improvements in eating habits (DeBar et al., 2011). On the other hand, interventions that target psychological well-being also provide benefit. For instance, longitudinal increases in self-esteem were associated with binge eating cessation in a population-based sample of adolescents (Goldschmidt et al. 2014). Relatedly, treatments that result in successful weight loss have also been found to reduce symptoms of depression and improve self-esteem (for a review, see Blaine, Rodman, & Newman, 2007). Thus, there may be a reciprocal relation between psychological well-being and weight and eating patterns, such that improvements in one may lead to improvements in the other.

Cognitive Behavior Therapy (CBT)

CBT for binge eating is comprised of behavioral techniques addressing nutrition (i.e., self-monitoring of food intake, education on regular and consistent eating, cue exposure) and cognitive skills ([identifying triggers to binge eating and learning new coping skills] Fairburn, Wilson, & Schleimer, 1993). Decaluwé and Braet (2005) found that Fairburn, Cooper, and Cooper's (1986) cognitive-behavioral model of bulimic symptoms, including binge eating, applied well to a sample of children and adolescents, ages 10–16 years. This suggests that models based on adult samples may also apply to youth, and therefore, treatments designed for adults may be effectively adapted for younger populations.

Research examining the effectiveness of CBT with children and adolescents with LOC eating has found evidence that it may be effective in reducing binge episodes. In particular, Braet, Tanghe, Veerle, Moens, and Rosseel (2004) developed a CBT approach for treatment-seeking obese children in an inpatient unit. Their approach did not include dieting, but instead focused on healthy eating, exercise, self-regulating, and planning. They found that after treatment, BED diagnoses were reduced from 37% to 5%, with sustained improvement at 14-month follow-up.

Recently, cognitive-behavioral treatments have been adapted for use on the internet. This can make treatment more cost-effective, accessible to patients, and acceptable to those who otherwise would not attend treatment. Jones et al. (2008) tested a similar 16-week, cognitive-behavioral internet program targeting binge eating; the program resulted in significant reduction in both weight and eating pathology, including binge eating (Jones et al., 2008).

Cue Exposure Exposure techniques have been implicated in CBT treatment, as the craving and eating behaviors of binge eaters are theorized to be cue-controlled (Jansen, 1998). Exposing patients to "trigger foods" that they typically find difficult to resist allows patients to concentrate on the food cues (i.e., sights, smells, opening wrappers) without consuming the foods. These cues will often trigger strong cravings and a desire to engage in binge eating, but after time these cravings will be extinguished.

Boutelle et al. (2011) evaluated two novel treatments to target overeating in children – cue exposure and an appetite awareness intervention. They found that both were effective in significantly reducing the frequency of binge eating, by helping children become more aware of their internal hunger signals, rather than focus on the external food cues.

The effectiveness of cue exposure techniques may depend on the age and developmental stage of the child. Braet et al. (2004) found that that young children may benefit from the immediate effects of extinction that result from cue exposure. Older children, due to their increased cognitive capacities, may benefit more from cue exposure supplemented with cognitive therapy (Braet et al., 2004). Therefore, clinicians should be sensitive to the age and cognitive abilities of their patients when planning treatment.

Guided Self-Help Facets of CBT have been used to develop guided self-help programs (GSH) for disordered eating. One GSH program in particular was written at an eighth-grade reading level and thus may be used in adolescent samples (Schmidt et al., 2007). In GSH, patients use a workbook under supervision of a clinician. These workbooks are typically comprised of elements of CBT, such as psychoeducation, thought monitoring, problem-solving, and goal-setting. Evidence suggests CBT-based GSH is more effective in producing binge abstinence than family-based treatments in adolescents with BN or EDNOS (DeBar et al., 2011; Schmidt et al., 2007).

Family-Based Treatment (FBT)

In the treatment of eating disorders in youth, parents are regarded as a major agent of change (Golan & Crow, 2004). For instance, Munsch et al. (2008) found that joint child-parent and parent-only treatments are equally effective in child weight control. While LOC was a treatment target, the authors did not report on frequency of specific eating behaviors; thus, it is unknown whether these treatments reduced LOC eating. When administered to adolescents, family-based treatments were shown to be more effective than a control treatment (supportive psychotherapy) at reducing bulimic symptoms including binge eating (Le Grange, Crosby, Rathouz, & Leventhal, 2007). Murdoch et al. (2011) also found a decrease in binge eating behaviors and food preoccupation after a 6-month, family-based treatment for obesity.

Interpersonal Psychotherapy (IPT)

The interpersonal model of BED (Wilfley et al., 2007) focuses on how deficits in social functioning contribute to low self-esteem and negative affect, which often precipitate binge eating. In support of this model, Elliot and colleagues

(2010) found that social problems in children and adolescents aged 8–17 were positively related to LOC eating, and this relationship was mediated by negative affect. IPT shows comparable efficacy to CBT (Robin, Gilroy, & Dennis, 1998; Wilson & Shafran, 2005). A randomized controlled trial showed that adolescent girls at risk for weight gain exhibited decreased LOC eating after a 12-week IPT program compared to a control group that underwent health education (Tanofsky-Kraff et al., 2010).

Behavioral Weight-Loss Treatment

Binge eating in youth often co-occurs with overweight and obesity (Allen et al., 2008; Tanofsky-Kraff, 2008). Therefore, behavioral weight-loss treatments – which emphasize healthy and regulated eating and exercise – may be a helpful means of reducing binge eating symptoms. However, this position is controversial, because some prior work has shown that weight-loss approaches can lead to more frequent binge eating and other disordered eating symptoms (Keys, Brozek, Henschel, Mickelson, & Taylor, 1950; Neumark-Sztainer et al. 2006). Furthermore, youth who endorse binge eating may not respond well to weight-loss treatment and have poorer long-term outcomes (Wildes et al., 2010).

Despite evidence that behaviors related to weight loss (e.g., caloric restriction) may result in binge eating, there is evidence that suggests that behavioral weight-loss programs under physician supervision can be effective without increasing disordered eating. Family-based behavioral weight-loss treatment resulted in significant weight reductions for both parents and children (Epstein, Paluch, Saelen, Ernst, & Wilfley, 2001). Children did not show significant increases in disordered eating, and their parents showed decreased binge eating.

Conclusions

We can draw several conclusions from our review of existing literature on LOC and binge eating in youth. First, LOC eating seems to be fairly common in samples of youth; future research must longitudinally determine for whom overeating and LOC eating develops into later BED. Second, developmental research suggests that subthreshold levels of LOC and binge eating may still be clinically relevant and associated with maladaptive outcomes. Incorporating these subthreshold LOC and binge eating behaviors into assessment, research, and treatment efforts is necessary. Third, much of the existing work on LOC and binge eating has been conducted in overweight/obese populations. Therefore, it will be necessary to more clearly separate risk for and effects of binge eating from those related to weight status. Finally, efforts testing the efficacy of approaches to treat LOC and binge eating in youth are in their infancy. It will be important to develop and longitudinally test treatments that have been tailored to the developmental stage of childhood and adolescent populations.

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Part IX

Externalizing Disorders: Neurodevelopmental Disorders

Autism Spectrum and Social Pragmatic Language Disorders

19

Andrew Livanis, Diana Almodovar, and Evan Skolnik

Part I: General Introduction

The story of the diagnostic entity named "autism" would make for a good dramatic series on Netflix. No other diagnostic category has caused so much controversy over the course of its history. Speculations regarding the etiology and definitions of autism have caused the public uproar and challenged the media to find convenient solutions and more importantly, villains and scapegoats.

On one end of the spectrum of speculative etiologies, we have the concept of "refrigerator moms" (Malott, 2004), an idea that seemed to captivate everyone in the medical and psychological community in the 1950s and 1960s, despite scientific evidence that clearly refuted this idea. Although no data exists to support the "refrigerator mom" proposition, the authors of the chapter are sure that countless families were torn apart by this downright silly idea. More recently we have seen the mantra of "vaccinations cause autism," an idea so dangerous that the British Medical Society saw fit to revoke the medical license of the person that presented the idea backed with falsified data (Burns, 2010). One would believe that the idea would have died with this disgrace, but it has found new life in an almost zombie-like fashion and has been championed by countless media figures.

The status of "autism" or more recently *autism spectrum disorder* (ASD) as a diagnostic construct also adds to the drama. For a long time, autism was considered a component of childhood schizophrenia, and it was only in the last 35 years that it has enjoyed its own category, distinct and different from other diagnoses. Recent revisions to the DSM diagnostic criteria, where certain categories such as *Asperger disorder* and *childhood disintegrative disorder* have been eliminated, have caused a maelstrom of debate and argument among the psychological, psychiatric, and parent advocate communities as a whole (Lord & Bishop, 2015).

Currently, ASD is categorized by the DSM-5 (American Psychiatric Association [APA], 2013) as a neurodevelopmental disorder, a condition that manifests at some point during a child's development and impacts multiple areas of life functioning. In order for a child to be diagnosed

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with ASD, she must (a) show deficits in social communication and social interaction in several contexts and (b) demonstrate restricted, repetitive patterns of behavior (RRBs).

For the first criterion to be satisfied (social-communicative deficits), children must demonstrate deficits in (a) social-emotional reciprocity, (b) nonverbal communicative behaviors, and (c) relationship management, to the point where portions of the child's life are disrupted. For the second criterion to be satisfied (RRB), children must demonstrate deficits in two of the following four areas: (a) stereotyped or repetitive movements, object manipulations, and/or speech; (b) insistence on "sameness"; (c) restrictive or fixated interests; and (d) unusual reactivity to sensory input, which cause significant life impairments. The domains and their symptom list are provided in Table 19.1.

Some researchers indicated concerns that the revision to the DSM-5 diagnostic algorithm might leave some children who went previously diagnosed (under the DSM-IV criteria) without a diagnosis (Kulage, Smaldone, & Cohn, 2014), although Lord and Bishop (2015) reviewed some analyses that suggested that few if any children identified by the DSM-IV would be left without any diagnosis under the DSM-5 criteria.

Table 19.1 DSM-5 autism spectrum disorder domain and symptom list

Social communication domain *Must demonstrate all of the following symptoms*	Restricted, repetitive patterns of behavior *Must demonstrate two out of the four symptoms*
Social-emotional reciprocity	Stereotyped/repetitive movements/object manipulations/speech
Nonverbal communication	Insistence on sameness
Relationship management	Restrictive or fixated interests
	Unusual reactivities to sensory input

Note: The symptoms must be present in early developmental periods (but may not become fully manifested until later in life). Symptoms must cause clinically significant impairments in important areas of current functioning

Professionals in the field continued to express concerns that children with social communication issues who do not present with any history or presence of RRBs would not be diagnosable. Hopefully the proposal of a new diagnostic category, *social communication (pragmatic) disorder* (SCD), will help practitioners deal with this population of children.

The intent of this chapter is to present the reader with an introduction to the DSM-5 criteria of ASD along with a review of other epidemiological data highlighting the nature, course, and treatment of the disorder. In addition, the authors will introduce and discuss the construct and diagnostic entity of SCD as well.

Autism Spectrum Disorder

For a very long time, researchers, practitioners, and parent advocate groups have almost reflexively used the word "spectrum" when referring to individuals who met the criteria for autistic disorder. Often the term "on the spectrum" was used as a synonym to imply that someone qualified for a diagnosis of autistic, Asperger, or pervasive developmental disorders. It has only been since the advent of the fifth edition of the *Diagnostic and Statistical Manual for Mental Disorders* (DSM-5; American Psychiatric Association, 2013) that the "spectrum" was formally incorporated into the official diagnostic nomenclature.

History of the Diagnosis and in the DSMs

Although the term was used previously in a colloquial manner, Lorna Wing (1996) was the first to coin the term *autistic spectrum disorders* in the research literature, albeit without extensive elaboration as to the makeup of the spectrum in her piece. Wing's notion of *autistic spectrum disorders* hypothesized that the many pervasive developmental disorders could be placed on a unidimensional axis, which ranged from the most severe to the least severe. On one end of her spectrum, there were a group of children that Leo

Kanner had described almost 50 years earlier, who presented with a group of severe symptoms that he named *infantile autism*. On the other end of Wing's spectrum, we had children whom Hans Asperger had described (just a few years after Kanner's paper) that presented a syndrome he named *autistic psychopathy* who spoke like small adults (Pearce, 2005).

Kanner's (1943) paper described a group of children who shared a group of features that we now identify as classic autistic behavior: preference to be alone than with others (in some cases bordering on not registering others at all), a resistance to changes in the environment, and disturbances in speech which led to idiosyncratic modes of communication. He named this syndrome *autism*, a term that he borrowed from a 1911 paper by Bleuler who had used it in reference to the rigid, inflexible, and self-referent thought patterns seen in individuals with schizophrenia (Volkmar et al., 2005). To Kanner, these children showed similar patterns to individuals with schizophrenia, although as Volkmar et al. (2005) remind us, the *autism* that described Kanner's group is a result of these children's failure to develop appropriate modes of interacting with the world and not a break from reality as was seen in schizophrenia. Unfortunately, it would take considerable time for Kanner and professionals in the greater psychological and psychiatric community to come to this realization, and the constructs of schizophrenia and autism were to be blurred for quite some time.

Kanner also described the parents of his case studies, whom he identified as intelligent, although they preferred to deal with ideas rather than people. In a follow-up presentation in 1955, Kanner (along with his colleague, Eisenberg) described many of these parents as "successfully autistic" implying that they displayed some of the same issues as their children, albeit to a much lesser degree (Baker, 2013). While Kanner's speculation regarding the parents of his sample was meant to be speculative, it was unfortunately a starting point for a variety of horrific ideas and practices put forth by the psychological community. Coupled with conceptual issues blurring the line between the *infantile autism* syndrome that Kanner described and schizophrenia, the psychoanalytic community latched onto a purely nurture-based etiology of autism. Children's upbringing was blamed for the presence of the *infantile autism*, and parents were clearly implicated. Specifically, these children's environments were described as cold, unnurturing, and lacking love that lead directly to the presentation of autistic-like symptoms.

Part of this problem was due to Kanner's conceptualization of the disorder. In his original 1943 paper, Kanner hypothesized that autism had a genetic etiology, although in his 1955 presentation with Eisenberg, he highlighted the psychogenic causes for the disorder. From that point and on, he flirted with interactional causal factors in the disorders; until finally in 1971, he reverted back to his original position and held firm for the rest of his career. While Kanner never used the term *refrigerator mother* (Baker, 2013), his shifts in opinion throughout his career left the door open for many to make the idea of cold uncaring parents (specifically mothers) as the cause of autism to be accepted by the medical and psychological communities as a whole as well as the public. While there are many psychoanalysts who used the model of a cold uncaring mother as the basis for their "treatments," the most famous is Bruno Bettelheim, who served as director of the Sonia Shankman Orthogenic School at the University of Chicago.

Dr. Bettelheim claimed that he had a PhD in the philosophy of education, when in reality he had taken his doctorate in art history and had taken only a handful of coursework in psychology (Grossman, 1990). Despite his lack of experience, he launched into his position as director and began to outline treatments that emphasized the long-term care of children with autism and other disabilities away from their parents. Many of the children who resided there have recently come forward to discuss the psychological and physical abuse that was inflicted on them at the Orthogenic School (Pollack, 1997), much of it by Bettelheim himself. Fortunately, the publication of copious scientific data supporting the genetic etiology of autism has since put the refrigerator mom theory as well as Bettelheim's treatments to

rest in the United States. On his deathbed, Bettelheim said that he did not want to be forgotten (Grossman, 1990); perhaps the psychological community should not forget what happens when the scientific community fails to respond swiftly to charlatans.

While Kanner wrestled with the intricacies of his *infantile autism* construct, an Austrian pediatrician named Hans Asperger published a paper in 1944 where he described a group of children between the ages of 4 and 11 who displayed adequate to above average cognitive abilities but evidenced idiosyncrasies in verbal communication (including nonverbal communication issues), social deficits which manifested in problems and behavior concerns with their peers, and a poor sense of proprioception (Klin, McPartland, & Volkmar, 2005). Even though Asperger and Kanner were not originally aware of each other's work, they both independently borrowed the term *autism* from Bleuler. Unlike Kanner, Asperger did take time to clearly distinguish his syndrome from schizophrenia. Since Asperger's writings were in German, they were not available to an English language audience until the 1970s and it was not until Lorna Wing published her paper in 1981 did the scientific community begin to look into Asperger syndrome more systematically.

The DSMs I–IV and Autism Spectrum Disorder

The first and second editions of the DSM sought to establish and standardize the nosology and nomenclature that psychiatrists used. There was little effort to conduct any research on the labels that were inserted into each edition, as there was a reliance on capturing what currently was used by the medical community. As such, many of the diagnoses were based on psychoanalytic theories that focused extensively on disordered parent-child interactions. It was in this atmosphere that the term *autism* was subsumed into descriptions of symptoms for schizophrenia. For example, in the DSM-II, the diagnosis *childhood schizophrenia* was described as a "condition that could be manifested by autistic, atypical, and withdrawn behavior" (APA, 1968). As a result, it is difficult to reliably evaluate any of the autism literature prior to the publication of the DSM-III. For example, some of Lovaas' early seminal work was conducted with groups of children who were, at the time, diagnosed with *childhood schizophrenia*, although in reading some of the descriptions of these children's symptoms, it appears that many of them qualified for a diagnosis of autism spectrum disorder (Lovaas, Freitag, Gold, & Kassorla, 1965).

It was in 1980 with the publication of the DSM-III (and in 1987 with the publication of the DSM-III-R) that scientific evidence was included as a factor when evaluating the validity of diagnostic categories. The DSM-III included many features that improved the diagnosis of autistic symptomology significantly over the last two versions of the manual. *Infantile autism* became a disorder of its own and was defined as a pervasive developmental disorder as distinct from schizophrenia; also the terms *autism* and *autistic* were removed from the definitions for schizophrenia. The DSM-III symptoms for infantile autism were arranged into three domains: minimal responsiveness to other people, communication impairments, and unconventional responses to the environment. The DSM-III-R also introduced the concept of an algorithm that required that diagnosticians identify 8 of the 16 symptoms listed in order for a child to qualify for a diagnosis of *infantile autism*. Finally, there was some appreciation for those children who displayed features of infantile autism, but did not display the minimal number of symptoms established by the algorithm (8/16) but still evidenced impairments socially or academically. The diagnosis of pervasive developmental disorder, not otherwise specified (PDD-NOS), was introduced for that subgroup.

The DSM-IV was released in 1994, and it refined the three symptom domains so that they reflected deficits in socialization, communication, and restricted and repetitive stereotyped behaviors. In addition, the fourth edition refined the algorithmic model of diagnosis, whereby six symptoms needed to be manifested, but at least two needed to come from the socialization domain, one from the communication domain and one from the repetitive

behaviors domain. The DSM-IV was the first to include Asperger disorder as a diagnosis, which consisted of impairments in socialization and restricted and repetitive stereotyped behaviors. In addition to Asperger disorder, three other pervasive developmental disorders were included as well – childhood disintegrative disorder and Rett's disorder – and the diagnosis of PDD-NOS was retained.

The DSM-5 and Autism Spectrum Disorder

The fifth edition of the DSM (2013) has done away with both the umbrella term pervasive developmental disorder (PDD) and the various subtypes included in that category. Unfortunately, the PDDs that were outlined to explore subtyping issues in previous editions of the DSM seemed to cause confusion among professionals and the public at large. Diagnoses of Asperger disorder and PDD-NOS, for example, lacked adequate inter-rater reliability (Lord et al., 2012) and suffered from uncomfortably low reliability (Bennett et al., 2008). Furthermore, clinicians were not able to adequately differentially diagnose people diagnosed with Asperger disorder from those individuals with higher-functioning autistic disorder (Bennett et al., 2008). Due to the conceptual vagueness of the DSM-IV nosological structure, clinicians began to develop their own local criteria for differentiating PDD subtypes (Lord et al., 2012). Whether a child was diagnosed with Asperger disorder or PDD-NOS usually depended less on symptom presentation and more on the clinic or site where the child was evaluated, which defeats the purpose of a standardized diagnostic nomenclature.

Another major change was the move from three domains of symptoms in the DSM-IV to two domains in the DSM-5 (i.e., social-communicative impairments and RRBs). Such a move made sense as much of the factor analytic research presented in assessment instruments suggested that a two-factor solution was more robust than a three-factor solution (Goldstein & Naglieri, 2010; Rutter, Le Couteur, & Lord, 2003; Lord, Rutter, DiLavore, & Risi, 1999) in explaining the variance of these diagnostic instruments and by implication, the diagnostic process as a whole. To a large part, combining the socialization and communication domains makes sense at a face validity level as it is difficult to imagine someone being social without performing some form of communication behavior and someone communicating without in some way being social.

More important were the organizational changes made among the symptom clusters within each domain. While the DSM-IV diagnosis of autistic disorder represented an improvement over the diagnoses offered in the DSM-III, it still amounted to a collection of symptoms grouped into three broad domains. Most of the symptoms listed in the DSM-IV generally applied to school-aged children, thus making the diagnosis of adults and infants or toddlers quite difficult. What the DSM-5 offers in its stead is a two-domain structure with symptoms that are arranged as conceptually bound spectra that elegantly lay out a range of possible symptoms that vary across age and setting.

For example, in the DSM-IV criterion, A(c) reads "a lack of spontaneous seeking to share enjoyment, interests or achievements with other people" (p. 60), while the DSM-5 criterion A1 reads: "Deficits in social-emotional reciprocity, ranging, for example, from abnormal social approach and failure of normal back and forth conversation; to reduced sharing of interests, emotions or affect; to failure to initiate or respond to social interactions" (p. 50). The DSM-IV symptom might have been a well-developed description but the DSM-5 symptom reads like a well-developed operational definition with good inclusive examples that could assist in the diagnosis of ASD in toddlers, school-aged children, or even the elderly.

Mapping autism onto a spectrum meant that a symptom once considered the hallmark of the definition of autism – failure to use verbal language – was not included in the DSM-5 diagnostic definition of ASD (Lord & Bishop, 2014). A few issues drove the removal of this symptom. Firstly, the developmental trajectory of many children on the autism spectrum reflected early language deficits that were remediated by intensive therapies. This improvement often

translated into diagnostic movement over the course of their lives – from autistic disorder to PDD-NOS. Removing the verbal language criterion served to decrease the need for these "diagnostic graduations." Secondly, the removal of Asperger disorder, a diagnosis that was applied to children with structurally typical (although pragmatically poor) language development, required that the spectrum be expanded to include children with this diagnosis within the spectrum. Finally, Lord and Bishop (2014) noted that language delays are a common symptom in many childhood profiles, so clinicians could account for language eccentricities with other language-based diagnoses (in much the same way that low cognitive abilities and deficient adaptive skills would be accounted for by the diagnosis of intellectual disabilities).

The spectrum that the DSM-5 conceptualizes is highlighted not by the severity of the issues the child experiences, but by how much assistance the environment must provide the individual (i.e., requiring "support," "substantial support," or "very substantial support"). These spectrum severity markers can serve to assist clinicians in determining the types of supports that these children will need as well as which level of intervention may work best.

Current Research on ASD

Symptom Presentation and Associated Features

Social-Communicative Deficits

The social-communicative domain is divided into three groups of symptoms. The first symptom grouping of the social-communicative domain involves deficits in social-emotional reciprocity, which is defined as an awareness of the emotional and/or interpersonal cues of others and the ability to respond appropriately to those cues in relation to the internal and external environment of others. The symptoms that would be seen that relate to this criterion refer to difficulties engaging with others and sharing feelings and thoughts, as well as responding to others' cues while in the environment. Table 19.2 highlights a partial symptom list for this criterion based on the DSM-5 text (2013) as well as a study conducted by Carrington et al. (2014) which attempted to map the *Diagnostic Interview for Social and Communication Disorders* (DISCO) onto the DSM-5 ASD criteria.

Clinicians familiar with the DSM-IV model of diagnosis will immediately recognize symptoms related to difficulties with socialization, understanding social cues, and the lack of reciprocity with language. Imitation is an important skill which develops naturally in typically developing children, but children on the autism spectrum imitate less frequently than their non-autistic peers, whether in spontaneous or prompted conditions (Davis & Carer, 2014). Indeed a good portion of preliminary applied behavior analytic interventions involve the direct instruction of imitation skills (Sundberg & Partington, 1998). Imitation is important for a variety of reasons, chief among those that it is the primary vehicle to

Table 19.2 DSM-5 continuum for Domain A1: Social-emotional reciprocity

Requiring very substantial support	Requiring support
No attempts to socialize with peers or any other groups (e.g., adults)	Few attempts to socialize that are often odd and poorly coordinated
No attempts to imitate others	Few attempts to imitate others
Lack of understanding of social cues	Difficulties with complex social cues, especially those pertaining to conversation
No attempts to seek comfort from or provide comfort to others	Minimal attempts to seek or provide comfort, often in an awkward or unconventional fashion
No interest in sharing ideas or toys or other areas of interest	Minimal attempts to share ideas, toys, or other areas of interest. At times these attempts to share are awkward
Few to no attempts to approach others socially, in many instances	Social approaches are often one-sided

Note: The symptoms on this list were adapted from the text of the DSM-5 (2013) and Carrington et al. (2014). These symptoms are meant to be illustrative but not exhaustive

learn new skills. For example, many conversational skills are developed simply by imitation and feedback loops that help children improve over time.

The second symptom grouping of the social-communicative domain involves deficits in nonverbal communication typically used when engaged in social interactions. These nonverbal communicative behaviors enhance or make the socialization "deeper" and "fuller." Eye contact, a symptom that has classically been linked to ASD since it was originally described by Kanner, may not be technically necessary for social interactions, but does increase the quality of the interaction as a whole and can increase the probability of future social interactions (Livanis, Solomon, & Ingram, 2007). The DSM-5 states that eye contact as a symptom is relative to cultural norms – perhaps in some cultures where direct eye contact is frowned upon, clinicians may look for body or head orientation toward the speaker. In addition to eye contact (or body orientation), facial features, speech intonation, and gestures may be lacking. For those children with better-developed language skills, difficulties coordinating verbal and nonverbal messages may be seen which can lead to a presentation of an awkward "body language."

In infants and toddlers, this criterion is manifested typically by deficits in joint attention. Joint attention is defined as the ability to establish a shared experience through eye contact, pointing and/or presenting objects to initiate a common interaction as well as the capacity to respond to such overtures by someone else's following eye gaze or sharing what they have presented. Children with poor joint attention skills have demonstrated poor language abilities and show difficulties acquiring language over time (Davis & Carer, 2014) (Table 19.3).

The third symptom grouping of the social-communicative domain involves deficits in relationship development and maintenance, when compared against others in the child's peer group, gender, and culture. In some instances, we might see minimal to no interest in peers. In young children this may be manifested through difficulties with play. Children with ASD tend to perform better when they engage in functional play (using a toy the way it is defined) as opposed to symbolic or pretend play (Kasari & Chang, 2014). For example, it would be easy for a child with ASD to play trucks with a toy truck, but it may be much more difficult for her to play with some sticks and pretend they were trucks. This functional play may not be diverse and may display extreme repetitive elements. What is important to assess when examining play is the level of interest (e.g., outright rejection of play with others or passive play interactions) or aggressive or disruptive approaches to play (Table 19.4).

Table 19.3 DSM-5 continuum for Domain A2: Nonverbal communication

Requiring very substantial support	Requiring support
Facial expressions not dependent on context	Social context determines few facial expressions
Lack of eye contact/body orientation toward speaker	Abnormal/minimal eye contact or body orientation toward speaker
No body language or gesturing when socially appropriated	Minimal or uncoordinated body language or gesturing
Developmental deficits in the development of nonverbal communication modalities	Failure to integrate verbal and nonverbal communication modalities
Poor joint attention	Speech intonation is deficient or nonexistent

Note: The symptoms on this continuum were adapted from the text of the DSM-5 (2013) and Carrington et al. (2014). These symptoms are meant to be illustrative but not exhaustive

Once children become older, there is a struggle to understand what the social norms are depending on the context, a notion that has been labelled as the *hidden curriculum* in some instances (Lee, 2010). The hidden curriculum refers to those rules that are not discretely taught but are "picked up" by most children. Unfortunately, children with ASD have difficulties identifying and internalizing these rules.

Restrictive or Repetitive Patterns of Behavior (RRBs)

In contrast to the DSM-5 requirements to the social communication domain, where all three symptoms *must* be met, the RRB domain includes

Table 19.4 DSM-5 continuum for Domain A3: Relationship development and maintenance

Requiring very substantial support	Requiring support
No understanding that behavior should be adjusted according to social context	Minimal understanding that context should determine behavior and in many instances, confusion as to how to change behavior
Play is either nonexistent or extremely limited. In many instances, repetitive functional play may be evident	Deficits in (or unwillingness to participate in) symbolic or pretend play. Some attempts at parallel play may be evident but they are awkward and uncoordinated
Limited to no attempts to engage in social interactions with peers. In some instances, there is no interest in interacting with peers	Some attempts to engage in social interactions with peers, but attempts are awkward and not integrated with the context. In some instances, there is no interest in interacting with peers

Note: The symptoms on this list were adapted from the text of the DSM-5 (2013) and Carrington et al. (2014). These symptoms are meant to be illustrative but not exhaustive

four groups of symptoms, and only two must be met. The first symptom involves a variety of stereotyped or repetitive behaviors such as simple stereotypies (i.e., behaviors that are stereotypical and repetitive in nature) like hand flapping, the repetitive use of objects (lining up toys, spinning objects), and/or echolalic responses. Echolalia can be immediate or delayed (repetition of a word or phrase mentioned previously). At times, students may display *scripting* behaviors, which involves the repetition of a script or series of interactions such as repeating the entire script from a cartoon or television show.

The second symptom includes those behaviors that are performed due to the child's insistence on sameness or the resistance to change. Typically children with ASD may become distressed when changes are imposed on the environment, from a change in food options to a fire drill in school. Some children who evidence symptoms from this criterion can be described as having difficulties with transitions at school or home which require a series of interventions to help deal with this. Alternatively, children with ASD may also evidence ritualistic verbal and nonverbal behaviors.

The third symptom refers to those highly restricted, fixated interests that are abnormal in intensity or focus. Some children may become attached to inanimate objects (the first author worked with a child that was attached to staple remover that he carried around with him) or may be preoccupied with unusual objects (the same child was also obsessed with the Metro-North Railroad system which connects New York City to its northern suburbs, even though he had never ridden that train). Some children display circumscribed or perseverative interests as well.

The fourth symptom refers to hyper- or hyporeactivity to sensory information. In some cases children may have an unusual reaction to sensory aspects of the environment, such as indifference to pain or illness, or adverse reactions to specific sounds or food textures. Excessive smelling or touching of various objects or parts of people (e.g., hair) may be evident as well.

Baranek, Little, Parham, Ausderau, and Sabatos-DeVito (2014) suggest that there may be several subtypes of RRBs, and certain symptoms from various domains appear to cluster together. Stereotypical behaviors, for example, seem to co-occur more frequently with hyperresponsiveness to environmental stimuli, inflexible routines, and other behaviors, as well as other compulsions, rituals, and behaviors related to maintaining "sameness." A possible second subtype may involve the hyporeactivity to sensory stimuli combined with sensory-seeking behaviors and behaviors related to the insistence on sameness. It is also important to note that hyporesponsiveness to sensory stimuli and sensory-seeking patterns has been associated with maladaptive behavior (Baker, Lane, Angley, & Young, 2008). While the research on the subtyping of RRBs is not well established, the diagnostician and clinician should keep them in mind when assessing and working with children with ASD.

Associated Features

It is important to highlight the effects that a child with ASD brings to the home in terms of stress to caregivers. In general, higher levels of child problem behaviors, regardless of the disability in question, are related to parental stress; however,

the mothers of children with ASD tend to report higher levels of distress than mothers of children with other developmental disabilities (Estes et al., 2009). Self-injurious behaviors appear to be the best predictors of parental stress (Konstantareas & Homatidis, 1989), while the evidence is inconclusive as to whether the level of adaptive daily living skills impacts parental stress levels (Estes et al., 2009; Tomanik, Harris, & Hawkins, 2004). However, we may be able to tentatively state that the more delays that the child with ASD presents, the higher the levels of parental distress that may be displayed.

Estimates of children dually diagnosed with ASD and intellectual disability (ID) have previously ranged into 50%, but current CDC estimates may suggest lower percentages (CDC, 2010). In their review of 11 sites throughout the United States, the CDC estimated that roughly 31% of children with ASD have overall intellectual scores that are below a standard score of 70, and 23% of children with ASD have intellectual scores that range from 71 to 85. (It should be noted that assessments of cognitive ability are not enough to diagnose an intellectual disability – the CDC sample did not include measures of adaptive functioning). There is some evidence to support that the presentation and prognostic outcomes of ASD tend to worsen as measures of intellectual functioning decrease (Matson, Tessa, Fodstad, & Boisjoli, 2009).

In addition, when samples of children with ASD are assessed using tests of cognitive ability, there tends to be a greater variability of scores on those instruments and a larger proportion of unexplained variance presented when compared to children without ASD or the normative sample (Nowell, Schanding, Kanne, & Goin-Kochel, 2015). Specifically, there appears to be a significant discrepancy between performance on verbal and nonverbal tasks, although the direction of such discrepancies is not constant (Black, Wallace, Sokoloff, & Kenworthy, 2009). Such discrepancies may be related to social communication issues.

Even when children with ASD score within the average range on assessments of cognitive ability, there appear to be significant gaps between their intellectual and measures of adaptive behavior functioning (APA, 2015). To a large degree, adaptive functioning may better predict prognostic outcomes in many cases of ASD (Kanne et al., 2011). In lower-functioning individuals with ASD, the development of functional language skills as well as effective nonverbal communication skills by age 5 can reliably predict better outcomes in adolescence and adulthood (Billstedt, Gillberg, & Gillberg, 2005). That finding has also been replicated in individuals with higher-functioning cognitive abilities – better prognoses are predicted by better adaptive functioning levels (Farley et al., 2009) – which raises important implications for the assessment and psychological treatment of ASD. It is unfortunate that schools do not seriously consider the adaptive skill deficits within their general curriculum for students with disabilities – their incessant focus on content leads them to view functional life skills as ancillary at best and interfering at worst (Ayers et al., 2011).

Some children with ASD tend to engage in aggressive, noncompliant, and/or self-injurious behaviors. The performance of aggressive behaviors may result in the further socially isolation of a child who is already struggling to understand and maintain relationships – peers may isolate the child in typical school settings, or the child may be enrolled in a restrictive educational facility which is often far from home (Luiselli, 2011). Self-injurious behaviors are one of the most baffling classes of behaviors that children with ASD may perform, often leading to hospitalizations at rates over seven times than seen in typically developing children (McDermott, Zhou, & Mann, 2008).

Deficits in motor imitation are captured by the social-emotional reciprocity (i.e., A1) criterion of the DSM-5 diagnostic algorithm, but there are a variety of other motor issues that children with ASD display. *Apraxia* (and its milder variant, *dyspraxia*), a condition where certain movements are restricted despite typical muscular development, is typically identified as a concomitant issue with children with ASD. When compared to children with ADHD, children with ASD showed deficits in basic motor development as well as gestural performance (Bodison & Mostofsky, 2014). In addition, children with

autism may tend to display significant problems with gait (Shetreat-Klein, Shinnar, & Rapin, 2014) and issues with using objects for their stated purpose (Dziuk et al., 2007) such as the use of a fork to eat or a toy to play.

Etiology

There is increasing consensus on the equifinality of ASD, which is to say ASD should be considered a disorder that has multiple causes (Happé & Ronald, 2008; Happé, Ronald, & Plomin, 2006). Researchers are also honing in on a variety of genetic factors as the main causal determinants of ASD as genetic factors may explain roughly 90% of the variance of the presence of ASD, although multiple genes and gene combinations are implicated in various pathways to ASD (Freitag, 2007).

It may be that ASD is caused by a genetic predisposition that is aggravated by some sort of environmental insult early in development, or in utero (Trottier, Srivastava, & Walker, 1999). It is beyond the purview of this chapter to describe all the potential environmental issues that may impact on the development of ASD, but a few warrant mention due to their scientific merits or their media controversy.

Beginning in utero (Gardener, Spiegelman, & Buka, 2009), bleeding and the maternal use of psychotropic medications during pregnancy are correlated with the incidence of autism, while prenatal exposure to rubella and cytomegalovirus can activate the mother's immune system and sympathetic nervous activity and increases the risk for the incidence of ASD. Conditions of the thyroid that decrease the secretion of thyroxine can also impact brain development during periods of development for the central nervous system (Román, 2007). Maternal stress has also played a disruptive role in brain development as well (Kinney, Munir, Crowley, & Miller, 2008).

A variety of postnatal environmental factors have been suggested, but there has been very little conclusive evidence to point the finger at one factor or another. While there are some data to support that children with ASD are more likely to have gastrointestinal (GI) symptoms than typical children (McElhanon, McCracken, Karpen, & Sharp, 2014), the current evidence suggests that GI issues are simply associated with ASD and that there is no causal relationship (MacDonald & Domizio, 2007). Some readers may recall that Andrew Wakefield attempted to coerce the medical and psychological communities to believe that some bowel disorders created conditions that allowed certain antibodies to be passed through the blood-brain barrier which contributes to brain dysfunction, thus contributing heavily to the development of ASD. Wakefield named this condition *autistic enterocolitis*, which he claimed allowed the thiomersal in the *measles*, *mumps*, and *rubella* (MMR) vaccine to pass through the blood-brain barrier and cause damage to various brain structures implicated in ASD.

Wakefield's hoax was found to be unreplicable, because Wakefield falsified the data (Burns, 2010; MacDonald & Domizio, 2007), in an attempt to obtain a patent for his own version of a MMR vaccine (Dyer, 2007). There is no link whatsoever between thiomersal, vaccinations, and the incidence of ASD (Doja, 2006). However, the public fear that swept the world has led to a variety of consequences. Currently, the United States currently has removed thiomersal content from its vaccinations; however, rates of ASD still continue to rise. Some parents who are still frightened from the hoax that Wakefield perpetrated are currently electing to not vaccinate their children leading to a host of other issues for infants, toddlers, and pregnant mothers who are exposed to a variety of viruses. It is critical to note that in 2004, the British Medical Council revoked Wakefield's medical license after he was found guilty of 30 charges including acting dishonestly and irresponsibly, circumventing ethics review boards, and receiving payment from lawyers who represented parents who believed that their children were harmed by MMR as well as several other counts (BBC, 2007, MMR doctor "paid children").

Comorbid Diagnoses

Assessing for the presence of emotional disorders (such as depressive and anxiety disorders) is difficult to perform. A core feature of ASD is the difficulty that children with the diagnosis have with sharing their thoughts and feelings, which would

make their self-report of depressive or anxious symptomology difficult to obtain. However, anxiety and depressive disorders are evident in the ASD population. In one recent meta-analysis, the prevalence of anxiety disorders in children with ASD was estimated to be roughly 40% (van Steensel, Bögels, & Perrin, 2011), and while there have been no meta-analyses of depression rates in ASD populations, Hollocks et al. (2014) indicate that rates may be as high as 70% among children with ASD. An increase in anxious and/or depressive symptomology is related to increases in functional and emotional impairments including adaptive functioning (Tureck, Matson, May, Whiting, & Davis, 2014), emotional regulation (Amstadter, 2008), positive academic outcomes (Duchesne, Vitaro, Larose, & Tremblay, 2008), as well as family, social, and emotional difficulties (Kovacs, Gatsonis, Paulauskas, & Richards, 1989).

It may be challenging to assess for anxious and depressive symptomology since, on the surface, several of the core features of the emotional disorders resemble symptoms or associated factors in ASD. There are considerable differences, however. In their sample of adults dually diagnosed with ASD and social anxiety disorder, Bejerot, Eriksson, and Mörtberg (2014) found that the quality of social anxiety was different from typically developing adults diagnosed with social anxiety disorder (SAD). Specifically, individuals with SAD had average or slightly less than average social skills but evaluated themselves rather poorly. Individuals with ASD and SAD presented significant social and communication deficits which gave rise to their anxiety. Interestingly, the authors speculate that a lack of self-evaluation skills may serve as a protective factor against social anxiety (and perhaps all other types of anxiety) – if individuals with ASD were not fully apprised of their issues, then perhaps they may not be as concerned about them, thus leading to less anxiety. While this is just conjecture on the authors' part, this may have some implications for the judicious application of interventions.

Similar to the emotional disorders, ASD and ADHD also share many symptoms and impairments (Mayes et al., 2011). For a long time, the diagnosis of pervasive developmental disorders precluded a diagnosis of ADHD even if symptoms of hyperactivity and inattention were noted. However, the current DSM-5 criteria suggest that the two disorders can exist comorbidly. Overall, children dually diagnosed with ADHD and ASD (when compared to children who were only diagnosed with ADHD) tend to show more daily living impairments and more inattentive and hyperactive symptoms and score lower on measures of cognitive ability. Furthermore, children who are dually diagnosed tended to display more intense and strongly expressed ASD symptomology (Craig et al., 2015).

Children with ASD demonstrate an increased prevalence of epilepsy and seizure disorders, reaching rates of up to 46% (El Achkar & Spence, 2015). Epilepsy in ASD appears to accompany the presence of ID and tends to appear more often in females than males (Amiet et al., 2008), perhaps due to the fact that ASD is expressed so much more strongly in females than males. The age of onset appears to be after 10–12 years of age although there are some instances where children will experience their first seizure prior to age 10 (Bolton et al., 2011). Currently there is no consensus as to how to screen for the presence of a seizure disorder in children with ASD, as many of the symptoms may appear to be similar (e.g., unresponsiveness, repetitive motions; El Achkar & Spence, 2015); therefore, the clinician should consult with a medical professional trained in diagnosing epilepsy to rule this out.

Course and Developmental Challenges

Parents will retrospectively report that they recognized something atypical about their child's development by the age of 24 months, although the age of recognition and/or formal diagnosis of ASD varies greatly (Chawarska et al., 2007). By 12 months, as many as 50% of parents may recall issues with impairments in social communication including a lack of response to name, failure to orient toward people's faces, lack of social smiling, reduced eye contact, lack of appropriate emotional facial expressions, and an inability to follow others' pointing and then later point to show items of interest to others (Zwaigenbaum, Bryson, &

Garon, 2013; De Giacomo & Fombonne, 1998). During the 12–24-month period, many of the same impairments in the social communication domain are evident, but children with ASD tend to also display reduced interest in peers and elect to be on their own.

The current research is not as clear when discussing RRBs within the first 24 months. While 12-month-old children with ASD displayed more elevated levels of repetitive motor actions than typically developing peers, their levels were equivalent to other developmentally disabled toddlers (Osterling & Dawson, 1994). Others have suggested that children with ASD did not differ from controls when looking at the quantity of typical object manipulation; however, the ASD samples tended to engage in significantly more atypical responses such as unusual visual and/or oral exploration or spinning, rotating, and moving objects in a repetitive fashion (Ozonoff et al., 2008).

The introduction of school beginning from preschool onward presents the first developmental and environmental challenge that the child with ASD (along with their family) will need to face. During this period, children move from a context where they are wholly dependent on their parents to arrange social interactions to a point where they can, over time, develop and maintain relationships with other students. Unfortunately, children with ASD tend to interact socially with their peers significantly less so than other children, which may lead to poorer quality socialization opportunities (Bauminger-Zviely, 2014). Indeed, what children with ASD do when socializing tends to be below age expectations – for example, their interactions are typically more awkward than their peers' and they evidence difficulties matching various nonverbal communicative behaviors (e.g., smiling, eye contact, and joint attention). The use of humor as well as the understanding of jokes is problematic. In addition, joint play behaviors may be challenging (Kasari & Chang, 2014). In fact, unstructured social activities may be quite difficult for the child with ASD to manage as they are asked to create or join into shared activities with their peers (Bauminger-Zviely, 2014). In other cases, RRB symptoms may necessitate alternative locations for some children (i.e., physical education and lunch may require a change in setting due to the traditionally noisy and chaotic nature of these environments).

The introduction of school also brings with it the demands and pressures of academic work. As the child with ASD progresses through the grades in school, the environment places ever more increasing demands for independent work. At this point many of these children may have already been diagnosed, but for many children, it is during this time that the vast majority of children are classified by their school systems and begin to receive services in one manner or another (Montes & Halterman, 2006). Often, it is the associated features of ASD and not its core symptoms that serve to be the most problematic to the management of academic work (Ashburner, Ziviani, & Rodger, 2010). For example, poor adaptive and/or intellectual functioning might necessitate specific curricula to teach skills directly in an ancillary fashion (when the diagnosis specifies a Level 1 severity level) or instead of portions of the curriculum that other children are exposed to (when the diagnosis specifies a Level 3 severity level). Self-injurious and aggressive behaviors may require significant environmental modifications in order to successfully deal with school during the day.

During adolescence, the child with ASD faces even more challenges. Upon entering middle and high school, children come into contact with teachers who become even more specialized in their topic. In elementary school, children may have interacted with a handful of teachers, while in middle and high school, children can have upward of eight to ten teachers, including in some instances paraprofessionals and teacher's aides. This greatly expands the network of individuals with whom the child with ASD needs to interact and can pose a series of issues.

Adolescents with ASD continue to experience the social-communicative issues that were evidenced in the past, but during this developmental period, they deal with puberty and the influx of bodily changes, issues that are difficult for any child to negotiate. Unfortunately, many adolescents and young adults do not receive appropriate

sex education instruction; parents and teachers are in favor of this idea, at least when polled, but often do not feel confident that they have the skills or knowledge to provide such educational experiences (Hellemans, Roeyers, Leplae, Dewaele, & Deboutte, 2010). Another issue is the sexual victimization of the child with ASD. Due to their limited ability to communicate well, Koegel, Detar, Fox, and Koegel (2014) indicate that they may be "perfect targets" for perpetrators of sexual misconduct. It should be noted that although girls are underrepresented in the ASD population (males are four times more likely to carry a diagnosis of ASD), they may tend to be more likely to show more intense intellectual and adaptive deficits, at least when clinic samples are compared (APA, 2013), which placed them in increased danger of becoming a victim of sexual misconduct. The implementation of well-developed sex education curricula however could counter these issues by teaching general safety and accurate reports skills.

Dealing with school-related issues may be difficult to manage at this point without direct support from parents and/or the school system. The trend toward the expectation of more independent work continues throughout middle and high school. Regardless of the levels of severity that the student evidences, many children will require some form of instruction to help them better develop their adaptive behavior skills (Turygin, 2014). One advantage of attending large high schools is the variety of after-school clubs that are available, some of which may match up with some of the interests that the teen with ASD has had since they were very young. Among those teens dually diagnosed with ASD and anxiety disorder, we tend to see an increase in behavioral avoidance and anxiety in social situations which may also cause some difficulties (Bejerot et al., 2014).

Treatment

Typing in "autism treatment" into a Google search will give you over 68 million web pages. It can be overwhelming for parents as well as clinicians to separate good interventions from "noise." While it is beyond the purview of this chapter to discuss each and every specific intervention, it would be helpful to review some heuristics that can help to select evidence-based treatments.

The National Autism Council (NAC) is a not-for-profit organization whose mission is to disseminate evidence-based information regarding the treatment of ASD. The NAC publishes a variety of literature reviews which categorize interventions into three broad categories: *established*, or the "gold-standard" interventions that carry with them a wealth of data; *emerging*, or those interventions that appear promising but simply do not have enough data to support them yet; and *unestablished*, or those interventions that either have no evidence to support them or can be harmful to children and families (National Autism Center, 2009). The NAC reports also work on identifying interactions noted in the literature, specifically age x intervention and ASD subtype x intervention (most of the reports were published before the DSM-5 was released). Currently there is a report that is due out in 2015 and it will be interesting to see if they follow the DSM-5 criteria.

Most of the interventions are based on the principles and procedures of applied behavior analysis. For example, 10 out of the 11 interventions identified by the NAC's National Standards Project, Phase 1 report (National Autism Center, 2009), are based on behavioral principles and procedures. Typically most parents and clinicians will assert that applied behavior analytic services are meant only for lower-functioning ASD children; however, the principles of behavior analysis are meant to be constants that are expressed as procedures to fit the needs and developmental levels of children. For example, the use of activity schedules is one intervention that has been identified by the NAC as an *established* treatment. Activity schedules are based on the principles of task analysis (breaking large tasks down into smaller, more workable steps) and chaining (reconnecting those steps to provide a workable sequence so that one can actualize a larger goal). With children who evidence Level 3 severity, we may see simple pictorial schedules, with perhaps no more than two or three steps that are provided to them during specific points in the day. Children with Level 1 severity levels may be taught to develop their own

schedules that may be text-based on a sheet of paper or phone/tablet.

As a further example, the first author has also used the delivery of reinforcement which was also identified as an *established* treatment with children with a variety of severity levels. At Level 3, many children required the delivery of food or toys immediately after almost each and every correct action that was performed. At Level 1, children were taught to identify appropriate actions and deliver positive self-statements to themselves. In addition, children with Level 1 severity ASD were asked to participate in a group of their peers and take some time each day to verbally reflect to on at least three positive actions that they had undertaken during the day and deliver positive self-statements (as well as hear it from other members of the group). The principle of reinforcement (i.e., certain environmental events will increase appropriate actions) ranged from a controlled delivery of specific concrete items at Level 3 to the instruction of a methodology to allow children deliver praise to themselves and to seek it out from others at Level 1. The applied behavior analytic principle remained the same, but its expression as a procedure varied depending on the level of severity and needs of the child.

Prognosis and Outcomes

While there has been some discussion regarding a "cure" for autism, especially from practitioners of complementary and alternative medicines (Treatment – Centers for Disease Control, http://www.cdc.gov/ncbddd/autism/treatment.html), ASD should be conceptualized as a chronic condition. The core deficits in the two domains will continue throughout the individual's childhood, adolescence, and adulthood, but the expression of those symptoms become less severe over time when compared to themselves (Howlin, 2006). Data suggest that over time, children with more severe needs and greater quantities and intensities of symptom expression tend to have poorer overall life outcomes (Howlin, Goode, Hutton, & Rutter, 2004).

Age of formal identification and the beginning of treatment appear to be related to a positive prognosis, with early identification and treatment involving evidence-based applied behavior analytic components signaling better outcomes (Chawarska et al., 2007). While there may be a desire to lump educational approaches of different orientations together to create unique interventions, there is evidence to suggest that these "eclectic" approaches are inferior to programs based on the principles and procedures of applied behavior analysis (Magiati, Tay, & Howlin, 2014). These applied behavior analytic services typically focus on the development of adaptive behavior skills, and their value may be in assisting in the development of a basic functional communication repertoire by 6 years, which is an important prognostic indicator (Howlin, Mawhood, & Rutter, 2000).

In addition, the presence of other concomitant conditions such as intellectual disability, language disorders, ADHD, or emotional issues can lead to poorer outcomes due to either the increase in the quantity of issues displayed or in some cases due to the increase in the severity of symptom expression. Therefore the early identification of such comorbid diagnoses and concomitant conditions is critical so that early treatments can be implemented to improve outcomes.

Social (Pragmatic) Communication Disorder

The DSM-5 introduced the diagnosis of SCD because there were some concerns that without it, there would be groups of children who displayed significant social communication issues (but no RRBs) that would not receive a diagnosis under the DSM-5 criteria for ASD and would not be eligible for services (Gibson, Adams, Lockton, & Green, 2013). Like ASD, SCD is categorized as a neurodevelopmental disorder, but is in a different subgroup which is named *communication disorders* (ASD forms its own subgroup under neurodevelopmental disorders; APA, 2013).

As a diagnostic entity, SCD is in its infancy, and there are very few studies that have examined this construct as a diagnosis. As of yet, there are no structured standardized instruments that can

assess this diagnostic construct. Researchers are also not clear as to the course, etiology, and its developmental trajectories over time. Due to the dearth of research, there has been a lot of "borrowing" that has occurred. For example, since there is a significant body of research on related disorders such as ASD and the role of pragmatics in language impairment, Norbury and Holloway (2013) have suggested that perhaps this may assist in the provision of a framework for its assessment, etiology, and course.

History of the Diagnosis

Over the past 20 years, there have been some issues that have emerged with the use of the DSM-IV diagnosis of pervasive developmental disorders (APA, 2000), specifically, the diagnosis of PDD-NOS. The diagnostic description of PDD-NOS appeared to be vague and described a condition which was named atypical autism in previous versions of the DSM. PDD-NOS referred to groups of symptom expressions that did not meet the criteria for autistic disorder due to atypical or subthreshold symptomology or a later age of onset. As was noted previously, such vague descriptions ran counter to everything that the DSM-III and IV attempted to do – offer clear-cut evidence-based descriptions of various disorders. As a result, diagnosticians interpreted this mysterious paragraph in a variety of ways (Lord et al., 2012), and we were left with a group of diagnosed children who demonstrated wildly discrepant behaviors.

Some reports indicated that children diagnosed with PDD-NOS tended to show fewer and at times no RRB symptoms (Maenner et al., 2014; McPartland, Reichow, & Volkmar, 2012), and their argument was that the DSM-5 diagnosis needed to accommodate for this "subtype." Lord and Bishop (2015) countered that the core features of ASD were a combination of social communication and RRB deficits, and those children who did not display both issues did not qualify for a diagnosis of ASD. There was no denying that these children evidenced significant difficulties which caused impairments in their daily lives, and if these children were to need a diagnosis, then a different construct is needed to be developed.

While the diagnostic debate continued, the research community has been looking seriously at those children demonstrate social-communicative deficits but not RRBs. This literature focuses on children with specific language impairment (SLI), which is defined by language impairment in the absence of frank motor, neurological, cognitive, sensory, and behavioral deficits (Leonard, 2014). There appears to be some disagreement, however, as to whether children with pragmatic deficits constitute a distinct subgroup of SLI children (Rapin & Allen, 1983) or whether pragmatic deficits represent a symptom of a relatively heterogeneous group of children with language delays (Brinton, Fujiki, & Sonnenberg, 1988; Craig & Evans, 1993). Yet some others attribute the decreased frequency and efficiency of pragmatic behaviors in children with SLI to be secondary to other linguistic deficits. For instance, children who present with difficulties in language comprehension, or difficulties in the production of structural aspects of language, such as phonology and syntax, may naturally be less inclined to socially engage others and demonstrate appropriate social skills (Bishop, Bishop, & Leonard, 2000; Fujiki & Brinton, 1993).

Bishop et al. (2000) in their comprehensive review highlighted studies that revealed a subgroup of children with SLI who demonstrate deficits primarily in the areas of language content (semantics) and use (pragmatics). These symptoms presented by this subgroup were initially referred to as *semantic-pragmatic deficit disorder* and was characterized by semantic deficits expressed through word-finding difficulty, atypical word choices, and comprehension deficits, with their phonological and syntactic skills intact. In addition, these children demonstrated a breakdown in communication on the discourse level and evidenced difficulties in topic initiation and maintenance. Rapin (1996) originally used *semantic-pragmatic deficit disorder* to describe deficits of this nature in other disorders (such as autism or some syndromes), while Bishop et al. used it to identify a specific subgroup of children

with SLI. In addition, studies examining pragmatic competence in children identified as having SLI found that a small subgroup of these children, despite requiring special services in school, scored within an average range on standardized test scores, yet were rated by their teachers as having severe communicative and social skills deficits (Conti-Ramsden, Crutchley & Botting, 1997).

Bishop et al. (2000) argued that a more appropriate label for this subgroup would be *pragmatic language impairment* (PLI). The characteristics of PLI as a group are largely consistent with the diagnosis of SCD in the DSM-5 (Adams et al., 2012). Gibson et al. (2013) distinguished the definition of SCD as outlined in the DSM-5 as compared to the research on children with PLI, in that children are viewed as falling somewhere on a spectrum, with SLI being on one end and autism on the other. PLI is not characterized as discretely as SCD is defined in the DSM-5, as its own distinct category. One defining characteristic separating PLI from ASD is the lack of RRBs in children with PLI. In addition, children with PLI demonstrate greater difficulties with peer interactions than children with SLI; however, these difficulties are less pervasive than what has been found in children with ASD (Gibson et al., 2013). Given these similarities, previous research identifying PLI as a disorder with characteristics distinctive from SLI and autism, it has been suggested that children whose profiles are consistent with SLI could fall into the category of SCD (Gibson et al., 2013).

In a recent editorial, Tager-Flusberg (2013) questioned the need for a category entirely for social communication deficits. She proposed that it is more appropriate to identify these deficits as features of other disorders (Tager-Flusberg, 2013). However, Lord and Bishop (2015) note that language-disordered symptoms are a part of so many disorders that it makes sense to have them separated into their own category in the DSM-5, to increase sensitivity and specificity in diagnostic decision-making. Nonetheless, the initial concept of SCD came from the observation that significant social communication and/or pragmatic language impairments have been displayed in children who do not meet diagnostic criteria for ASD (Bishop & Norbury, 2002). In addition, the research on PLI (formerly semantic-pragmatic disorder) over the past 30 years provides a useful construct upon which future research on SCD and as well as application by clinicians can move forward.

Current Research on SCDs

Symptom Presentation and Associated Features

The DSM outlines four diagnostic criteria for SCD, all of which must be present in order for a diagnosis to be met. The first of these criteria specifies the presence of deficits in the social use of verbal and nonverbal communication. These, specifically, must include deficits in utilizing conversation for social purposes, adjusting the style of communication to the context, understanding and following conversational rules, and nonliteral and inferential communication. The social deficits in question must result in anxiety about speaking or participating in a variety of domains.

Third, the social communication deficits must have an onset in the early developmental period. Early communicative behaviors begin to emerge from birth, with a newborn showing some recognition for a caregiver's voice, typically signified through the newborn's gaze pattern. Early red flags of atypical development of communication skills include but are not limited to no eye contact or eye gaze, lack of or reduced responsiveness to auditory or visual stimuli in the environment, delayed emergences of communicative gestures, and late onset of first words. The final criteria specify that the symptoms are not attributable to another condition such as a medical or neurological condition, low abilities in speech or language, intellectual or developmental delays, or autism spectrum disorder.

Differential Diagnoses

Currently there are no structured mean to distinguish ASD from SCD. To accurately diagnose SCD, practitioners must determine that a client's

social and communication symptoms must not be better explained by ASD, which, unlike SCD, includes restricted, repetitive patterns of behavior, interests, or activities. Before a diagnosis is made, practitioners should consider that stereotyped language, a common feature of SLI (Bishop & Norbury, 2002), has been added to the definition of restricted and repetitive behaviors in the DSM-5 criteria for ASD.

It should be noted that children diagnosed with SCD may display slightly but nonsignificantly elevated levels of RRBs compared to peers (Reisinger, Cornish, & Fombonne, 2011). As such, it will be important for the clinician to evaluate whether the RRBs cause clinically significant impairment in daily living conditions. A subset of individuals that would have formerly been diagnosed with a DSM-IV diagnosis of a pervasive developmental disorder (i.e., autistic disorder, Asperger disorder, PDD-NOS, childhood disintegrative disorder) may likely display such subthreshold RRBs, and a diagnosis of SCD should be considered.

However, practitioners must be careful not to automatically assign all individuals previously diagnosed with PDD-NOS with SCD. As was noted earlier in the chapter, most individuals who were diagnosed with a pervasive developmental disorder under the DSM-IV criteria would qualify for a diagnosis of ASD under the DSM-5 criteria. Kim et al. (2014) found that over two thirds of the individuals diagnosed with PDD-NOS under DSM-IV criteria would meet criteria for ASD under DSM-5 criteria. The vast majority of the remaining individuals (about a third PDD-NOS participants) did meet criteria for SCD, with the rationale being that these individuals did exhibit RRBs but were not at the level, rate, or intensity to impact functioning in one or more areas. The remainder of the PDD-NOS cases actually fit criteria for an attention-deficit or anxiety-related pathology, which again reflect Lord and Bishop's (2015) claim that many of the pervasive developmental disorder symptom criteria were too open to interpretation and depended more on the clinician performing the task of diagnosis as opposed to the symptoms expressed.

Etiology, Course, and Developmental Challenges

It is becoming clearer that there is a genetic predisposition for social-communicative deficits and related disorders (Skuse & Gallagher, 2011; Pourcain et al., 2010). There is an observable genetic link between a common allele for autism spectrum disorder and social communication deficits, most notably stereotyped conversation and decreased ability to understand pragmatics (Pourcain et al., 2010).

While those with family members with clinical social impairments appear to be at a greater risk for SCD, the severity of these disorders may not be solely determined by genetics. Applied behavior analysis at the early intervention level is an effective means of improving symptoms of ASD and its comorbid deficits (Matson & Konst, 2013). Thus, along with ASD, children with SCD may show great resilience when they receive verbal, behaviorally geared, early intervention. In addition to direct treatment, fostering a positive outlook in the individuals with this disorder will likely foster resilience. Keyfitz et al. (2013) found self-efficacy to be predictive of resilience.

Assessment and Treatment

Checklists have shown to accurately identify aspects of pragmatic deficits that can be missed by traditional cognitive testing. The *Children's Communication Checklist* (CCC) is one scale that has been shown to do this accurately in elementary populations (Bishop, 1998). Social communication skills can be assessed with the *Social and Communication Disorders Checklist* (SCDC). The SCDC has been shown to be a valid brief screener for social reciprocity and verbal/nonverbal communication (Skuse, Mandy, & Scourfield, 2005). The structured observation format of the *Autism Diagnostic Observation Schedule* (ADOS) has been shown to measure conversation skills. This scale allows for qualitative observation of unusual communication behaviors and is more appropriate for adolescents and adults (Lord, Rutter, DiLa- vore, & Risi, 2001). Thus, there are a variety of instruments to target and conceptualize the needs of individuals with SCD. Additionally, these tools are not only

useful for assessment but also for progress monitoring. Norbury et al. (2013) hypothesized that diagnostic instruments are able to demonstrate improvements in symptoms.

There is a clear lack of cohesive theories regarding the treatment of SCD (Gerber et al., 2012; Norbury et al., 2013), although there is some evidence that measurable changes in social communication behavior have been observed when intense and manualized intervention is utilized (Adams et al., 2012). The treatment of SCD may be aimed at helping these children increase communication skills, make improvements in social relationships, and prevent outcomes such as disruptive behavior and social withdrawal. Children with SCD command years of support especially as they get older, and the expectation for social competence increases (Norbury et al., 2013).

The developmental individual-difference, relationship-based model (Greenspan, Wieder, & Hollander, 2007) teaches how to improve relationships; social skills; meaningful, spontaneous use of language; and understand human development. This approach has been shown some evidence that it might increase spontaneous communication and back-and-forth communication (Pajareya & Nopmaneejumruslers, 2011; Greenspan & Wieder, 1997). The data suggest that a program that structures and reinforces basic back-and-forth interactions, silly and anticipatory play, social problem solving, and utilizing words and symbols to communicate ideas (Greenspan et al., 2007) may have promise for effectiveness for the SCD population. The National Autism Center's *National Standards Project* (2009) lists DIR as an *emerging* treatment for ASD, which suggests that its evidence is limited when applied to children with ASD. However, in the report, it indicates that one of the populations that received this intervention was children who were diagnosed with PDD-NOS. It is quite possible that DIR might actually be an intervention that is better suited to be delivered to children with SCD, and its effects all along were minimized when a population of children with ASD (in the form of PDD-NOS) were added to their samples.

Interventions should target language processing issues as well as related difficulties such as reading comprehension proficiency related to sentence-level oral comprehension (Westby, 2011). One such intervention is the *Social Communication Intervention Project* which demonstrated significant treatment effects in the areas of conversational competence, pragmatic skill, social communication, and teacher ratings of classroom learning, after 20 sessions of intensive sessions with a speech-language pathologist (Adams et al., 2012). This intervention focuses on three factors: social understanding, language pragmatics, and language processing through the use of graphic organizers, checklists, imaginative play, art projects depicting social situations and various emotions, and routine development.

Final Comments

While the construct of autism has caused a considerable amount of controversy over its lifetime, the last 20 years have provided the most research on autism than at any other point in history. Such research has allowed us to better understand its nature, course, and prognosis, as well as help researchers and diagnosticians set the boundaries that separate what is and what is not considered to be autism spectrum disorder.

The DSM-5 is by no means a perfect document, but when examined in terms of autism spectrum disorder, it is the best diagnostic description that is currently available. The DSM-5 guidelines provide for a streamlined set of symptoms in an attempt to increase the rates of specificity (the ability of the criteria to correctly identify those children with ASD) and sensitivity (the ability of the criteria to correctly identify those children without ASD; Lord & Bishop, 2015). It represents the first attempt to legitimately define symptomology vis-a-vis a spectrum. Finally, it has done away with the multitude of diagnostic categories that are associated with the diagnosis of ASD (e.g., Asperger disorder, childhood disintegrative disorder, PDD-NOS), which served to confuse practitioners and ultimately led to their implicit

abandonment of the DSM as an anchor for diagnosis.

As such, the DSM-5 criteria should be carefully examined and evaluated over time. For instance, what of the estimated 33% of individuals who do not qualify for a diagnosis of ASD (Kim et al., 2014)? While reports suggest that these individuals may qualify for other newly introduced diagnoses such as SCD, it is still not clear whether SCD is a valid diagnosis and further research into this area needs to be explored. Furthermore, it appears that subtyping RRB's syndromes or patterns might be useful as they may help in the development of treatment options for children with behavioral difficulties and issues.

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Attention-Deficit/Hyperactivity Disorder

20

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Overview of ADHD and History in the DSMs

Attention-deficit/hyperactivity disorder (ADHD) is a neurodevelopmental disorder comprised of clinically significant inattention and/or hyperactivity (American Psychiatric Association [APA], 2013). ADHD is a relatively common disorder that affects approximately 5% of the population (Willcutt, 2012), differentially affects males more than females at a 2:1 or more ratio (Owens, Cardoos, & Hinshaw, 2015), and is associated with significant impairment in academic, social, and/or occupational functioning (APA, 2013). Symptoms and impairment are typically manifested early in life and are chronic across the lifespan for most individuals (Barkley, 2015). Thus, it is critically important to diagnose this condition as early as possible and deliver effective interventions on a consistent basis across home, school, and community settings.

This chapter will first provide a brief history of the diagnostic criteria for ADHD with a detailed description of current DSM-5 criteria. Next, a multimodal assessment protocol will be delineated with an emphasis on documenting symptoms and impairment thereby connecting assessment with the development of effective home- and school-based interventions. Third, the accumulating evidence indicating that ADHD is caused by differences in neurophysiological structures and processes that have genetic origins will be described briefly. Fourth, a detailed overview of symptom presentation, course of the disorder, and developmental challenges will be provided. Fifth, sociodemographic trends in ADHD identification as well as associated impairments and comorbid disorders are discussed. Sixth, treatment for ADHD is described including pharmacotherapy and psychosocial interventions. Finally, the chapter concludes with an overview of the prognosis and expected outcomes for children with ADHD.

History of ADHD

Descriptions of children displaying the symptomatic behaviors of ADHD have been found going back to the 1700s (Barkley, 2015). Still (1902) is credited with the first medical description of the disorder with an emphasis on deficits in "moral control." As a result of an encephalitis epidemic in the early 1900s, children were noted to exhibit postencephalitic behavior disorder that was manifested by high levels of impulsivity, motor activity, and compromised attention and

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memory skills (Strecker & Ebaugh, 1924). Subsequently, children displaying inattentive, impulsive, and/or disruptive behavior were believed to have experienced brain damage even if the structural deficits were too minimal to be detected (Strauss & Lehtinen, 1947). Continued interest in possible neurobiological underpinnings for the constellation of ADHD symptoms led investigators to identify such children as displaying hyperkinetic impulse disorder (Laufer, Denhoff, & Solomons, 1957). The emphasis on the motor activity component of ADHD was exemplified by the inclusion of hyperkinetic reaction of childhood disorder in the second edition of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM; APA, 1968).

Over the course of the 1970s and 1980s, there was increasing recognition of the central role of attention difficulties in the symptomatic expression of ADHD (Barkley, 2015). Virginia Douglas's model of attention deficits (Douglas & Peters, 1979) was particularly influential in the eventual coining of the term "attention deficit disorder" or ADD as part of the third edition of the DSM (APA, 1980). The DSM-III criteria for ADHD were notable for several reasons. First, the criteria involved a shift from hyperactivity to inattention as the central deficit for the disorder. Second, specific criteria were provided that included (a) descriptions of symptomatic behaviors across three domains (i.e., inattention, impulsivity, and hyperactivity), (b) thresholds for requisite number and chronicity (i.e., 6 months) of symptoms as well as age of onset (i.e., 7 years old), (c) requirement for symptoms to be associated with impairment in functioning across two or more settings, and (d) description of disorders to be ruled out (e.g., autism). Third, two subtypes (i.e., ADD with hyperactivity and ADD without hyperactivity) were identified based on the presence or absence of hyperactive symptoms. The publication of DSM-III criteria was a watershed moment for a veritable explosion in research and clinical work focused on ADHD.

Subsequent versions of the DSM have involved substantive changes in several criteria; however, the core symptoms of inattention and hyperactivity-impulsivity have remained. In the absence of compelling research that supported subtypes, the DSM-III-R (APA, 1987) grouped 18 symptoms into one list. This was also the first time the disorder was referred to as ADHD. Based on factor analytic studies and large sample field trials, the DSM-IV (APA, 1994) grouped the 18 symptoms into two categories (i.e., inattention and hyperactivity-impulsivity). Further, on the basis of a growing research base supporting the existence of an inattentive subtype (Carlson & Mann, 2000), the DSM-IV included three subtypes based on symptom thresholds in each domain. Specifically, ADHD predominantly inattentive type was evident when significant symptoms of inattention were present and significant symptoms of hyperactivity-impulsivity were not. ADHD predominantly hyperactive-impulsive type was designated when significant symptoms of hyperactivity-impulsivity were present and significant symptoms of inattention were not. Finally, ADHD combined type was identified when significant symptoms in both domains were evident.

Current Diagnostic Criteria for ADHD

ADHD is currently conceptualized as a neurodevelopmental disorder characterized by the display of developmentally inappropriate frequencies of inattention and/or hyperactivity-impulsivity (APA, 2013). Significant symptoms in one or both of these behavioral domains are associated with functional impairment wherein children with ADHD demonstrate difficulties with delaying responding to the environment, developing self-control, and maintaining consistent work performance over the course of time (APA, 2013; Barkley, 2015).

The behaviors, or "symptoms," comprising ADHD according to DSM-5 criteria (APA, 2013) include difficulties with attention (e.g., often has difficulty sustaining attention in tasks or play activities) and hyperactivity-impulsivity (e.g., often has difficulty waiting his or her turn). To be considered symptoms of ADHD, some of the behaviors must have been initially exhibited in childhood (i.e., prior to age 12) and must be chronically displayed across two or more settings (APA, 2013). An individual must be reported to exhibit at least six of the nine

inattention symptoms and/or at least six of the nine hyperactive-impulsive symptoms. For older adolescents and adults over the age of 17, at least five symptoms in at least one of these domains must be exhibited. The ADHD diagnosis is usually determined by establishing the developmental deviance and pervasiveness of symptoms. It also is important to rule out alternative causes for the individual's inattention, impulsivity, and motor restlessness including poor academic instruction and management practices; gross neurological, sensory, motor, or language impairment; intellectual disability; or severe emotional disturbance (Barkley, 2015).

One of the most important changes to the diagnostic criteria for ADHD is the move from subtypes to symptom presentations. This change was based on longitudinal studies indicating that ADHD subtype status was fluid across development (i.e., that an individual's subtype designation was likely to change over time) (Willcutt, Nigg, Pennington, Solanto, & Rohde, 2012). Thus, according to the DSM-5, there are three presentations of ADHD (APA, 2013). The combined presentation describes individuals who exhibit the critical number of symptoms of both inattention and hyperactivity-impulsivity for a minimum of 6 months. This is the "classic" variant of ADHD that has been studied widely in the literature and is the most problematic presentation. ADHD predominantly inattentive presentation (previous terms have included "undifferentiated attention deficit disorder" and "attention deficit disorder (ADD) without hyperactivity") is diagnosed in those children exhibiting at least six of the nine inattention symptoms and fewer than six hyperactive-impulsive behaviors. Finally, ADHD predominantly hyperactive/impulsive presentation is diagnosed for those children who display at least six of the nine hyperactive-impulsive symptoms but less than six inattention symptoms.

Assessment of ADHD

A behavioral assessment approach is employed in the evaluation of ADHD wherein multiple methods of data collection are utilized across informants and settings (see Anastopoulos & Shelton, 2001; Barkley, 2015; Pelham, Fabiano, & Massetti, 2005). In particular, emphasis is placed upon obtaining reliable information regarding a child's behavior from parents and teachers as well as from firsthand observations of individual performance. Thus, the major components of the evaluation include interviews with the child's parent(s) and teacher(s), questionnaires completed by parents and teachers, and observations of child behavior across multiple settings and under varied task conditions. Although many of these same procedures are used when evaluating adolescents, some modifications (e.g., inclusion of self-report measures) are necessary to maintain the reliability and validity of the assessment data.

Interviews with the parent(s), teacher(s), and child are conducted to determine the presence or absence of symptoms of various DSM-5 disorders (including ADHD) as well as to identify historical and/or current factors possibly serving to maintain identified problem behaviors. Behavior rating scales (e.g., *Behavior Assessment Scale for Children-2*, *Conners Rating Scales*) completed by the child's parent(s) and teacher(s) provide data that establish the severity of ADHD-related behaviors as well as behaviors associated with other disorders (e.g., conduct problems, anxiety, mood disturbance) relative to a normative sample. To supplement parent and teacher report, several direct measures of student behavior are used. The child's behavior is observed across settings (e.g., classroom and playground) on several occasions to establish the frequency and/or duration of various target behaviors. Behavioral frequencies are usually compared to those displayed by several of the student's classmates to determine the deviance of the referred child's behavior. Finally, permanent products of the child's behavior (e.g., academic productivity and accuracy, quality of desk organization) can be collected and/or examined. Although each of these techniques is limited in some manner, when used in a multimodal assessment package, a system of "checks and balances" develops such that the drawbacks of any single measure are balanced by data obtained through other means (Anastopoulos & Shelton, 2001; Barkley, 2015).

Self-report questionnaires completed by children and adolescents have also been developed to assess symptoms of ADHD and other emotion and behavior disorders (e.g., Conners, 2008). Examples include the *Youth Self-Report* (Achenbach & Rescorla, 2001), the *Youth Inventory-4* (Gadow et al., 2002), and the *Self-Report of Personality for the Behavior Assessment System for Children-2* (BASC-2; Reynolds & Kamphaus, 2004). Given long-standing concerns that elementary and middle school-aged children with disruptive behavior disorders are typically poor reporters of their own behavior (Landau, Milich, & Widiger, 1991) and may overestimate their academic and social competence (i.e., positive illusory bias; e.g., Hoza, Pelham, Dobbs, Owens, & Pillow, 2002), self-report ratings, particularly of disruptive behavior, academic functioning, and social skills, should be interpreted with caution. Alternatively, growing evidence suggests adolescents with behavior disorders may be able to provide information that may aid in both diagnostic (Conners, 2008) and treatment (Smith, Pelham, Gnagy, Molina, & Evans, 2000) decisions. Also, self-report data are important to assess covert areas of functioning (e.g., depressive symptoms) and to engender adolescent cooperation with the evaluation and treatment process.

Two important components of the assessment of ADHD include establishing that symptoms lead to clinically significant impairment and ruling out alternative hypotheses for what appear to be ADHD symptoms. With regard to assessing for possible impairment, it is particularly important to include measures evaluating academic and social functioning. Academic functioning can be assessed using teacher ratings of academic performance (e.g., *Academic Competency Evaluation Scale*) and/or organization skills (e.g., *Children's Organizational Skills Scales*), direct evaluation of specific academic skills (e.g., curriculum-based measurement), and/or evaluation of permanent products (e.g., calculating percentage of assigned work completed correctly). In terms of ruling out alternative hypotheses for apparent symptoms of ADHD, clinicians should consider several possibilities. First, inattentive and/or hyperactive-impulsive behaviors could result from the frustrations encountered due to academic difficulties. If, for instance, the child begins to exhibit ADHD symptoms later in childhood after several years of learning difficulties or only exhibits problem behaviors during academic instruction in weaker subject areas, then this possibility should be considered. A second possibility is that the child is encountering emotional and/or adjustment difficulties (e.g., secondary to parental divorce or death in family) that have led to inattentive, impulsive, and/or restless behaviors. Third, changes in attention and activity level may actually be symptoms of other disorders (e.g., anxiety or mood disorders) and not represent ADHD. Finally, poor or inconsistent academic instruction and/or behavior management practices are other possible causes of apparent ADHD symptoms. This hypothesis should be explored whenever assessment data are inconsistent across sources and settings—for example, parents and teachers disagree about the severity and frequency of ADHD symptoms.

Etiology of ADHD

There is no apparent single etiological mechanism or "cause" of ADHD. Rather, ADHD symptomatology may result from a variety of and interaction among causal factors (Barkley, 2015; Nigg, 2006; Nigg & Barkley, 2014). Most studies examining the etiology of ADHD are correlational. Thus, caution is warranted in attributing causal status to identified variables. With this caveat in mind, substantial empirical evidence indicates that within-child variables, such as neurobiological factors and hereditary influences, account for at least 70–80% of the variance in ADHD symptoms (Barkley, 2015; Nigg & Barkley, 2014). More specifically, research has shown possible etiological roles for structural differences in the brain (e.g., abnormalities in the frontal-striatal network; Schweitzer et al., 2000), exposure to toxins (e.g., cigarette smoking during pregnancy; Mick, Biederman, Faraone, Sayer, & Kleinman, 2002), and genes related to the dopaminergic system (Banaschewski, Becker, Scherag, Franke, & Coghill, 2010). It is important to note that there is growing consensus that the heritability of ADHD is related to multiple

genes each of which may contribute a small percentage of the variance in symptomatic presentation (Nigg, 2006; Nigg & Barkley, 2014).

Environmental influences (e.g., family stress, poor parental disciplinary practices) do not play as large a causal role as neurobiological variables, but appear to modulate the severity of ADHD symptoms (Barkley, 2015; Nigg, 2006). In addition, environmental experiences may moderate genetic contributions (Martel et al., 2011). That is, ADHD symptoms may be associated with gene-environment interactions. Thus, the preponderance of evidence indicates that multiple neurobiological factors may predispose children to exhibiting higher rates of impulsivity and motor activity along with shorter than average attention spans compared to other children. Specifically, genetic factors and/or early exposure to a neurotoxin may alter the size and functioning of prefrontal and striatal neural networks that serve as planning and organization centers of the brain (Nigg, 2006). Furthermore, behavioral genetic studies have generally supported the notion of ADHD as a dimensional rather than a categorical disorder wherein everyone exhibits the symptomatic behaviors of this disorder on occasion. What separates individuals with ADHD from typically developing peers is that the former may be genetically predisposed (through neurobiological differences) to exhibit these behaviors at a significantly higher rate than others of the same age and gender. Despite the fact that neurobiological variables appear to be primary causal factors, environmental factors play a prominent role in the maintenance of ADHD symptoms. As discussed later in this chapter, interventions that involve the manipulation of environmental conditions can be quite effective in enhancing the functioning of children and adolescents with this disorder.

Symptom Presentation, Course of the Disorder, and Developmental Challenges

Across all ages, children and adolescents with ADHD display symptoms of inattention, impulsivity, and hyperactivity. Although these symptoms are pervasive and often problematic across time, the presentation of symptoms can vary as children progress through different ages and developmental stages. As such, the following section includes a description of children's common ADHD symptoms in preschool, elementary school, and secondary school through adolescence.

Preschool

Oftentimes, parents and teachers first notice a child's ADHD symptoms once the child enters preschool. Preschool is typically the first time in most children's lives in which they are expected to follow a set of rules that require sustained attention and self-control. This can be particularly difficult for children with ADHD, who may then "stand out" as having atypical behavior (Wolraich, 2006). In preschool, very young children with ADHD display more problem behaviors, poorer social skills, and more negative social behaviors than children without ADHD (DuPaul, McGoey, Eckert, & VanBrakle, 2001). Similar behaviors can also become apparent outside of the school setting, both during parent-directed activities (e.g., cleaning up a room) and in free play settings. Children with ADHD tend to display more frequent inappropriate and noncompliant behaviors toward their parents than children without ADHD at home and in other settings (DuPaul et al.).

Elementary School

Children who demonstrate ADHD symptoms at a very young age are likely to continue displaying similar inattentive, hyperactive, and impulsive behaviors as they progress through elementary school. This pattern of continuous symptom presentation exists across areas of impairment, including academic (e.g., lower rates of work completion), social (e.g., lower social preference by peers), health (e.g., higher rates of unintentional injury), and general adaptive functioning (Lahey, Pelham, Loney, Kipp, & Ehrhardt, 2004). ADHD symptoms are apparent in multiple settings and can be noticed by educators in the school

setting and parents at home and in other settings. Children with ADHD tend to score lower than children without ADHD on various indicators of academic performance, including achievement testing of reading/language, mathematics, and writing, and teacher ratings of academic competence and academic performance (McConaughy, Volpe, Antshel, Gordon, & Eiraldi, 2011). Elementary school children with ADHD also display social and emotional impairments; they often lack close friendships or positive relationships with family members, frequently due to difficulties with emotional regulation, which in turn may lead to high levels of conflict with family members and peers (Wehmeier, Schacht, & Barkley, 2009). Seemingly minor ADHD symptoms in very young children may become more problematic as the children exhibiting those symptoms progress through elementary and secondary school.

Secondary School

When children either show symptoms of or are diagnosed with ADHD in preschool or early elementary school, it is very likely that they will continue to be excessively hyperactive, impulsive, and/or inattentive as they reach adolescence. Depending on the definition of "recovery," it is estimated that somewhere between only 15% and 25% of children who are identified as having ADHD at a young age will be considered well adjusted once they reach their teenage years (Lee, Lahey, Owens, & Hinshaw, 2008; McGee, Partridge, Williams, & Silva, 1991). Interestingly, Hurtig, Ebeling, Taanila, Miettunen, and Smalley (2007) demonstrated that inattentive symptoms are likely to persist into adolescence, while hyperactive symptoms often fade, even for individuals who displayed a great deal of hyperactivity into childhood. Hurtig et al. describe adolescents with ADHD as having "dreamy symptoms," such as difficulty with organization, forgetfulness, or avoiding long or boring tasks. It is possible that adolescents actually internalize their hyperactivity, which results in feelings of internal restlessness as opposed to outward displays of disruptive behaviors. Finally, it is important to note that although symptoms may diminish as children grow older, a diagnosis of ADHD remains characterized by levels of attention, impulsivity, and/or activity-related symptoms that are significantly more severe that those of their peers.

Sociodemographic Trends in ADHD Identification

Presumably, the actual incidence of childhood ADHD is similar across various individual differences. Nevertheless, many assert that ADHD is a distinctly American phenomenon attributable to cultural factors (Faraone, Sergeant, Gillberg, & Biederman, 2003). Meta-analyses of domestic and international studies have consistently found the prevalence of ADHD in the United States to be comparable to other countries (Faraone et al., 2003; Polanczyk, de Lima, Horta, Biederman, & Rohde, 2007). One such analysis, which pooled 171,756 children from 102 studies, found only a minimal effect of geographic origin—studies in the USA found significantly higher rates of ADHD than studies in the Middle East or Africa but did not differ from those in South America, Asia, Europe, or Oceania (Polanczyk et al.). The prevalence of ADHD may be on the rise as more recently, Visser, Danielson, Bitsko, Holbrook, and Kogan (2014) found that approximately 11% of children and adolescents in the USA had ever received an ADHD diagnosis with 8.8% having a current diagnosis, based on parent report. These rates of ADHD diagnosis represent an increase of 42% between 2003 and 2011.

Despite global similarities in prevalence, several important diagnostic trends exist between groups of American children. First, boys are two times more likely than girls to receive an ADHD diagnosis (Visser et al., 2014). Second, African-American children are disproportionately identified as exhibiting high levels of ADHD symptoms, particularly through rating scales completed by teachers (DuPaul et al., 1997; Nolan, Gadow, & Sprafkin, 2001). Conversely, African and Hispanic-American children are less likely to be diagnosed by a physician even after controlling

for insurance coverage and income (Pastor & Reuben, 2005; Visser et al., 2014). Once diagnosed, African-American children with ADHD are less likely than their white counterparts to be prescribed medication by a physician (Pastor & Reuben, 2005). This inverse relationship between treatment and diagnosis among African Americans may be driven in part by disproportionate poverty. Froehlich and colleagues (2007) found that the poorest 20% of children are more than twice as likely as the wealthiest 20% to meet criteria for ADHD, but that they were only 33% as likely to regularly take medication for ADHD if they met criteria. Researchers and practitioners should consider potential biases in systems, measures, and informants used to identify and treat children with ADHD.

Associated Impairments

Academic

Academic impairments can be a major issue for children and adolescents with ADHD. From as young as preschool age, children with ADHD demonstrate difficulties with functioning both in terms of academic performance and behavioral functioning in academic settings. DuPaul et al. (2001) found that quantitative measures of these deficits for children with ADHD might be as far as two standard deviations below typical academic and behavior performance indicators for preschoolers. Further, young children with ADHD symptoms have been found to score lower on measures of reading, spelling, and math; this is especially apparent for children with inattentive symptoms (Massetti et al., 2008; Spira & Fischel, 2005). This is quite problematic as it shows that there is a huge gap in the academic skills of children with ADHD and their peers even from the very start of their education.

Scholastic struggles persist as children grow older and reach adolescence, with a moderate-to-large discrepancy in academic achievement between older children and adolescents relative to comparison peers (Frazier, Youngstrom, Glutting, & Watkins, 2007). That being said, Frazier et al., through a meta-analysis, also found a trend of decreasing academic impairment as students get older. This decreasing trend might be explained by the fact that those with the greatest degree of impairment either do not complete high school or attempt higher education. It is also possible that ADHD symptoms affect academic performance less as children grow older, either because the symptoms actually do decrease or because individuals better manage their inattention, hyperactivity, and impulsivity tendencies.

Additionally, executive functioning seems to play an important role in the academic outcomes of children with ADHD. Executive functioning behaviors that are related to academic performance include the ability to plan and follow through with tasks as well as organization of time and materials (Langberg, Dvorsky, & Evans, 2013). Children displaying ADHD symptoms are much more likely than their peers to show executive functioning deficits, and, in turn, those deficits are related to an increased risk of academic difficulties such as lower educational achievement and greater risk of grade retention (Biederman et al., 2004b). These issues with planning and organization can be particularly challenging when it comes to homework, including writing down homework assignments, staying on-task when working on homework, and actually completing the entire assignment (Langberg et al., 2011; Power, Werba, Watkins, Angelucci, & Eiraldi, 2006).

Vocational

About one in five adolescents between the age of 16 and 19 who attend school are also employed (Federal Interagency Forum on Child and Family Statistics, 2014). Despite this, virtually no research has examined the impact that ADHD has on occupational functioning during adolescence. Broadly speaking, adolescents with emotional and behavioral disabilities (including ADHD) have been found to overrate their job performance relative to ratings provided by supervisors (Carter & Wehby, 2003). Young adults with ADHD are significantly

more likely than their counterparts without ADHD to be both unemployed and not in postsecondary school (16.6% vs. 2.4%), and, among young adults receiving postsecondary education, students with ADHD are less likely to be engaged in employment (16.1% vs. 30.1%; Kuriyan, Pelham, Molina, Waschbusch, & Gnagy, 2013). Compared to young adults without ADHD, young adults with the disorder are significantly more likely to be fired and for employers to report ADHD-related symptoms while at work (Barkley, Fischer, Smallish, & Fletcher, 2006).

Social

As is the case for academic impairments in children with ADHD, social difficulties are apparent from a very young age. Preschoolers with ADHD symptoms tend to spend more time engaging in independent play activities and less time participating in group play than their peers without those symptoms (Alessandri, 1992). When they do become engaged in interactive play, they display more negative behaviors than children without ADHD, appear to lack awareness of their peers' feelings and needs during those interactions, and struggle to maintain two-way interactions that lead to prolonged engagement in peer play (Cordier, Bundy, Hocking, & Einfeld, 2010a, 2010b; Melnick & Hinshaw, 1996). These social interaction deficits are also evident during competitive games, during which children with ADHD are more likely to violate rules and focus on winning rather than on having an enjoyable experience with a peer (Melnick & Hinshaw; Normand et al., 2011). Also, children with ADHD tend to have close friends with relatively high levels of ADHD symptoms and oppositional behaviors, as well as fewer positive experiences and more problems in their friendships compared with children without ADHD (Normand et al.).

In addition to having difficulty in social interactions with peers, children with ADHD may also experience problems in family relationships. DuPaul et al. (2001) demonstrated these challenges through multiple assessment methods. In that study, parents of children with ADHD rated themselves as feeling more stressed and experiencing greater family dysfunction than parents of children without ADHD. Also, observations of parent-child interactions showed that the children with ADHD were more noncompliant and displayed more inappropriate behavior and both the parents and children exhibited more frequent negative behavior toward the other than in exchanges between parents and children without ADHD. Tripp, Shaughency, Langlands, and Mouat (2007) obtained similar findings and observed more engagement, more warmth, and more effective communication in families not including a child with ADHD.

Associated Comorbid Disorders

The high co-occurrence of ADHD with a number of other psychological disorders is well-documented. There is a particularly high comorbidity rate between ADHD and learning disabilities (DuPaul, Gormley, & Laracy, 2013; DuPaul & Stoner, 2014). Approximately 33% of students with ADHD also meet criteria for a learning disability, and approximately 38% of students with an LD meet criteria for ADHD (DuPaul et al., 2013). The direction of this relationship is unclear: it is both possible that, in some cases, students with an LD struggle to attend to material that is difficult to understand, while simultaneously, in other cases, symptoms associated with ADHD may prevent learning of a given topic. Regardless of directionality, there is evidence that ADHD and LD share underlying biological mechanisms (e.g., Willcutt, Pennington, Olson, Chhabildas, & Hulslander, 2005), and practitioners working with children and adolescents with ADHD should be prepared to assess for and address learning deficits.

ADHD also has high rates of comorbidity with other externalizing disorders, such as oppositional defiant disorder (ODD) and conduct disorder (CD). In fact, the comorbidity rate of ADHD with ODD and CD often exceeds 30% or 40% (Jensen, Martin, & Cantwell, 1997;

Newcorn, Halperin, Jensen, Abikoff, & Arnold, 2001). This high comorbidity rate may be particularly true among children with either combined or predominantly hyperactive/impulsive presentations of ADHD (Campbell & Ewing, 1990). Students with ADHD and comorbid CD or ODD (but no other comorbidity) may respond particularly well to treatments including medication (Jensen, Hinshaw, Kraemer, Lenora, & Newcorn, 2001). As such, practitioners and clinicians should be prepared to assess for the presence of disruptive behavior in children and adolescents with ADHD, as well as intervene with children and families in the case that such behaviors are present.

There also is significant evidence of substantial comorbidity between ADHD and various internalizing disorders, including anxiety and depression (Biederman, Newcorn, & Sprich, 1991; Spencer, Biederman, & Mick, 2007). For example, lifetime prevalence of depression in children with ADHD is estimated to be 45% by age 15 (Biederman, Faraone, Keenan, Benjamin, & Krifcher, 1992). Given that anxiety may present similar to ADHD (e.g., restlessness), clinicians should take appropriate steps to isolate the two diagnoses in assessment. Finally, there is some evidence that internalizing problems may actually *reduce* negative outcomes associated with ADHD symptoms: symptoms such as social withdrawal and anxiety may reduce the likelihood that children and adolescents with ADHD will engage in impulsive behavior or affiliate with peer groups involved with risky behavior (Masten, Roisman, Long, Burt, & Obradović, 2005).

Finally, there is substantial evidence that children with ADHD have an increased risk for other additional diagnoses. Most notably, children and adolescents with ADHD are more likely to be diagnosed with a substance or alcohol-related disorder (Kessler, Adler, Barkley, Biederman, & Conners, 2006) or to be diagnosed with Tourette's syndrome or tic disorder (Biederman et al., 1991). In particular, clinicians working with adolescents with ADHD should be vigilant for potential issues with substance and alcohol abuse and dependence.

Pharmacotherapy for ADHD

The prescription of psychotropic medication is the most common treatment for ADHD with approximately 70% of children diagnosed with ADHD receiving pharmacotherapy (Visser et al., 2014). Approximately 6.1% of children and adolescents (representing 3.5 million children) are treated with psychostimulant medications (e.g., methylphenidate, dextroamphetamine, mixed amphetamine compound) in the USA (Visser et al.). The highest rates (approximately 7% of the population) of stimulant treatment are for children between 5 and 14 years old (Zito et al., 2008). Psychostimulant medication use has grown steadily over the last few decades, particularly among preschool and secondary school populations (Visser et al.). The average duration of medication use is between 2 and 7 years, depending upon the age of the child (Safer & Zito, 2000). Further, more research has been conducted on the effects of stimulant medications on the functioning of children with ADHD than any other treatment modality for any childhood disorder (Connor, 2015).

Numerous studies have consistently demonstrated short-term enhancement of the behavioral, academic, and social functioning of the majority of children being treated with stimulant compounds. Several meta-analyses indicate that effects on behavioral and social functioning are in the moderate to large range with improvements over placebo conditions varying from 0.5 to more than 1.0 standard deviation units (Faraone & Buitelaar, 2010; Van der Oord, Prins, Oosterlaan, & Emmelkamp, 2008). Smaller effects in the range of 0.33 standard deviation units have been found for academic functioning (Van der Oord et al., 2008). Stimulants appear to exert greater effects on problems associated with ADHD symptoms (e.g., academic underachievement) when combined with other effective treatment approaches, such as behavior modification (MTA Cooperative Group, 1999).

The most frequent adverse side-effects associated with CNS stimulants include loss of appetite and insomnia, with less common side-effects such as headaches, stomachaches, and irritable mood occurring as medication effects are wearing off

(Connor, 2015). Some children with ADHD treated with methylphenidate may demonstrate constriction of cognitive functioning (i.e., overfocused behavior) that includes persistence at a task for an abnormally long period of time, a disregard for relevant peripheral stimuli, or an inability to shift cognitive set to adapt to situational demands (Solanto & Wender, 1989). Empirical investigations have failed to document the occurrence of this phenomenon at a group level of analysis; however, at least one study has indicated that children who are less hyperactive and who demonstrate better baseline performance on cognitive testing relative to other children with ADHD may exhibit overfocused performance as a result of methylphenidate use (Solanto, 2000). Stimulants, particularly dextroamphetamine, may also be associated with reduction in growth over time especially with respect to weight gain (Faraone, Biederman, Morley, & Spencer, 2008). In most cases, these adverse side-effects are lessened or eliminated by a reduction in dosage or a switch from one stimulant medication to a different stimulant compound (Connor 2015).

Because not all children and adolescents respond positively to stimulant medication and some individuals experience significant adverse side-effects, other psychotropic medications also are used to treat ADHD. Alternative medications include antidepressants (e.g., bupropion), antihypertensives (e.g., clonidine and guanfacine), and atomoxetine. Although all three types of compounds, particularly atomoxetine, have been found effective for reducing ADHD symptoms (Tanaka, Rohde, Jin, Feldman, & Upadhyaya, 2013), stimulants remain the medication of choice due to larger effects on symptoms and associated impairments as well as their relatively immediate behavioral effects (Connor, 2015).

Psychosocial Treatment for ADHD

Interventions for Teachers

Teachers may use interventions for their students with ADHD that either focus on academic performance, such as increasing work completion or accuracy, or classroom behavior, such as sitting still during instruction or raising a hand before speaking. The interventions with the strongest empirical support are those that involve modification of antecedent events (i.e., environmental stimuli that occur prior to a specific target behavior). Rathvon (2008) notes that these proactive teacher strategies have three defining characteristics: (a) they are preventive, meaning they work to minimize environmental cues for the undesired behavior before the behavior occurs; (b) they integrate behavior management and instruction into one comprehensive plan rather than treating them as two separate issues; and (c) they encourage desired behavior from the entire classroom instead of singling out one misbehaving student. Examples of antecedent interventions that are supported by research include increasing the degree of structure in instruction, integrating choice into assignments, presenting and reviewing material both orally and visually, increasing active engagement and responding during instruction, using explicit instructions, providing immediate and frequent feedback, and using peer tutoring to supplement teacher instruction (Raggi & Chronis, 2006).

Another option for an intervention that can be implemented by a teacher is a school-home note or daily report card system (Volpe & Fabiano, 2013). These communications between the teacher and parent(s) include feedback about the child's performance in and out of the classroom and rewards to reinforce desired behaviors. The effectiveness of school-home notes in increasing on-task behavior and academic productivity has been demonstrated in multiple studies (Jurbergs, Palcic, & Kelley, 2007; Kelley & McCain, 1995; McCain & Kelley, 1994). Those same studies found that school-home notes were equally effective with and without the inclusion of response cost, but also discovered that participants in those studies reported preferring for the school-home notes to include response cost.

Finally, teachers can participate in in-service trainings to become better informed about ADHD and how to effectively work with students with ADHD. A study by Jones and Chronis-Tuscano (2008) showed that teachers who took part in an

in-service training about working with students with ADHD were more knowledge able about ADHD after the training. Also, the special education teachers who participated in the training reported significantly increased use of behavior management techniques. This shows promise for the potential of improving educators' ability to serve students with ADHD in the classroom.

Intervention with Families

Parent behavior training is a well-established method for improving outcomes for children with ADHD. These interventions are provided to the parent, rather than the child, typically in a class-like format, and are designed to teach strategies that will help parents manage their child's behaviors with improved discipline strategies and parenting skills (Charach et al., 2013). One such program is *The Incredible Years*, a training program offered for children up to age 12 with disruptive behaviors along with their parents and teachers (Webster-Stratton, 1999). The parent training component of *The Incredible Years* teaches parents to promote healthy child development, strengthen the relationships they have with their child, and utilize positive discipline strategies. *The Incredible Years* has been found effective in reducing ADHD symptoms and disruptive behaviors (Jones, Daley, Hutchings, Bywater, & Eames, 2007).

Barkley (2002a) has developed a parent behavior training program geared toward reducing defiant behaviors while increasing rule following in children with ADHD. During training sessions, a therapist reviews the causes of defiant behavior, describes general strategies for reducing problematic behaviors, and teaches parents specific behavior management strategies that can be used at home and in school. The goal of Barkley's program is to provide parents with knowledge about how to help their children with ADHD that the parents can continue to apply after the class is complete to encourage long-term success.

Another training program for parents of children with disruptive behavior disorders is called the *Community Parent Education Program* (COPE; Cunningham, Bremner, Secord, & Harrison, 2009). There are two versions of COPE, one for parents of children ages 3–12 and another for parents of adolescents ages 13–18. Through COPE, parents learn skills to improve their relationships with their children and guide their children to learn about cooperation and problem solving (Cunningham et al.). Examples of skills discussed during COPE sessions include resolving conflict, managing transitions, and communication. The effectiveness of COPE has been demonstrated through research, particularly when the program is implemented in community settings (Cunningham, Bremner, & Boyle, 1995).

Parent-child interaction therapy (PCIT) is another intervention that may be effective in guiding parents to help their children with ADHD through parent-child interactions and relationship-building strategies. PCIT uses live coaching from a therapist to teach parents specific methods for interacting with their children through parent-directed and child-directed interactions (Eyberg, 1988). Studies of the outcomes of PCIT have shown that this type of intervention can reduce disruptive behaviors in children with ADHD (Wagner & McNeil, 2008).

Although the primary purpose of behavioral parent training programs is to improve the functioning of children with ADHD, research has also established that parent-focused interventions can be beneficial to the parents themselves as well. Anastopoulos et al. (1993) found that parents who participated in a parent training reported decreased parental stress and increased parental self-esteem following the completion of the intervention. The children of those same parents showed decreased symptom severity after the intervention. These findings are important because they demonstrate that parent-focused interventions not only help the target child, but the larger family system as well.

Direct Intervention with Children

There is a large body of literature evaluating the effectiveness of various different methods of directly intervening with a child or adolescent

with ADHD. As opposed to encouraging changes in behavior by altering the physical environment or changing the manner in which parents or teachers interact with the child, these interventions directly target the student's own skills or behaviors. DuPaul and Eckert (1997) compared several school-based interventions for ADHD, which they divided into three distinguishing categories: cognitive behavior, contingency management, and academic interventions. Broadly speaking, their meta-analysis found that classroom-based interventions are effective and that academic and contingency management interventions are particularly helpful in reducing disruptive classroom behavior exhibited by children with ADHD.

Other meta-analyses have found classroom-based strategies similarly effective (DuPaul, Eckert, & Vilardo, 2012; Stage & Quiroz, 1997). In particular, Stage and Quiroz (1997) found that response cost interventions and token economies for differential reinforcement were most effective at reducing disruptive behavior. Whereas response cost interventions involve taking tokens from students when they display a targeted behavior (where remaining tokens can be used to buy rewards), token economies involve providing tokens to students when they exhibit a target positive behavior. There also is evidence that self-monitoring and peer-monitoring interventions can be used to reduce disruptive behavior in school settings (Davies & Witte, 2000; Gureasko-Moore, DuPaul, & White, 2006).

Interventions designed to specifically remediate deficits in organizational skills have demonstrated efficacy in academic settings. These interventions might include guided practice in using a planner or training in organizing notebooks, binders, or other school materials. One example is an 8-week training designed by Langberg, Epstein, Urbanowicz, Simon, and Graham (2008), which focused on homework completion. As a function of training, students with ADHD exhibited significant improvements in completing homework, increases in GPA, and reductions in problem behavior around homework.

Several interventions designed for students with ADHD focus on attempting to reduce deficits in social skills commonly exhibited by children and adolescents with ADHD. These interventions may target a child's social difficulties through teaching the child basic skills needed to enter social situations, maintain interactions, and solve problems or conflicts (Sheridan, Dee, Morgan, McCormick, & Walker, 1996). Although some social skills interventions have shown promise, practitioners should be cautioned that, in general, these strategies have shown little generalizability of effects to other social situations (Evans, Axelrod, & Sapia, 2000; Gresham, Sugai, & Horner, 2001). Those hoping to implement an intervention for social skills should seek to create as naturalistic situations as possible, such as providing students with feedback immediately after observed social interactions.

Very limited research exists examining direct interventions for ADHD outside of a school setting—for example, home-based interventions generally target parent behavior. One example of a home-based direct intervention for ADHD is mindfulness training, which has been shown to have some efficacy in improving markers of attention (Zylowska, Ackerman, Yang, Futrell, & Horton, 2008). Alternatively, the research on using mindfulness training to reduce problems associated with ADHD is generally limited relative to other, behavioral strategies or mindfulness as applied to other outcomes.

Prognosis and Outcomes of Children with ADHD

Although the vast majority of research addressing ADHD has focused on children, many individuals diagnosed with ADHD will continue to meet diagnostic criteria for ADHD into adolescence and adulthood. The extent to which a diagnosis of ADHD will remit largely depends on the definition of remissions employed: among boys, as many as 60% of children with ADHD will no longer meet diagnostic criteria once in mid to late adolescence, although almost all (90%) will still have impairment associated with ADHD symptoms

(Biederman, Mick, & Faraone, 2000). A number of individual differences predict whether ADHD will persist through childhood and adolescence into adulthood: specifically, children with more severe ADHD symptoms are more likely to persist, as are children with comorbid diagnoses such as anxiety, CD, and ODD and children with maternal psychopathology (Biederman, Petty, Clarke, Lomedico, & Faraone, 2011). Additionally, the presentation of ADHD symptoms often changes over time: whereas among children there are significant differences between the manifestation of symptoms in males and females, these differences are no longer significant during adulthood (Biederman, Faraone, Monuteaux, Bober, & Cadogen, 2004a). One driving reason for this change may be the developmentally typical reduction in hyperactive symptoms (e.g., inability to stay seated) with age, as compared to the relative durability of inattentive and impulsive symptoms (Schmidt & Petermann, 2009).

ADHD has been linked to a number of negative outcomes. First, adolescents with ADHD have an increased likelihood of engaging in risky use of alcohol, tobacco, or other drugs as compared to their peers without ADHD, including abuse, addiction, and early initiation (Charach, Yeung, Climans, & Lillie, 2011; Molina & Pelham, 2003). Although characteristics associated with ADHD such as impulsivity are independently related to substance use among adolescents, adolescents with ADHD are especially at-risk for negative substance-related outcomes when they affiliate with deviant peer groups (Marshal, Molina, & Pelham, 2003). As such, clinicians working with adolescents with ADHD should not only be alert to potential use of drugs, alcohol, and tobacco even at a young age, but should consider the individual's peer group in assessing risk.

Second, youth with ADHD tend to underperform on various measures of academic achievement relative to their peers without ADHD (Frazier et al., 2007). A review of the vast literature examining the academic outcomes of children and adolescents with ADHD found that children and adolescents with ADHD are more likely to be retained, more likely to be disciplined at school, have lower grade point averages, are more likely to drop out of high school, and are less likely to attend college (Barkley, 2002b).

Thirdly, as discussed previously, ADHD is consistently associated with psychological comorbidity, and the presence of comorbidity tends to increase the likelihood of negative outcomes (Banaschewski et al., 2010; Kessler et al., 2006). Fourthly, children and adolescents tend to have lower performance on indicators of successful independent functioning. For example, adolescents with ADHD are more likely to fare poorly on various markers of driving competency (such as moving violations and accidents) and to be incarcerated relative to their same-age peers without ADHD (Barkley, Guevremont, Anastopoulos, DuPaul, & Shelton, 1993; Teplin, Abram, McClelland, Dulcan, & Mericle, 2002). Lastly, ADHD is associated with a number of social difficulties, including elevated conflict with parents, serious peer relationship problems, and affiliation with deviant peer groups (Barkley, 2002b). Unfortunately, there is no evidence that these deficits are offset with advantages—studies investigating the presence of potential advantages associated with ADHD, such as increased creativity, have generally failed to find positive differences favoring individuals with ADHD (e.g., Healey & Rucklidge, 2005).

Conclusion

ADHD is a chronic, relatively common neurodevelopmental disorder that is associated with significant impairment in academic, social, and/or occupational functioning. The conceptualization of the disorder has evolved over the years from an emphasis on putative underlying brain damage to a focus on hyperactivity and attention deficits. Consistent with factor analytic and field trial data, DSM-5 criteria include two symptom dimensions (inattention and hyperactivity-impulsivity) that are manifested from an early age. This chapter provided current information regarding the etiology, symptom presentation, assessment, and treatment of ADHD with a particular emphasis on evaluation and intervention directed to functional impairments. Although substantial research

supports the use of psychotropic and psychosocial treatments to reduce the frequency and severity of ADHD symptoms, more comprehensive and focused investigations are necessary to evaluate interventions that ameliorate educational and social functioning in order to circumvent the problematic long-term outcomes frequently associated with this disorder.

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Tourette's and Tic Disorders

21

Keith D. McGoldrick

Introduction

Abigail

Abigail is 7 years old. She has an unremarkable medical history with normal developmental progress. She is of above-average intelligence and neuropsychological abilities. Abigail's attention is on point. She is socially adept. However, at 5 years old, Abigail began displaying an involuntarily licking her lips and atypical eye blinking. Most recently, involuntary puckering of her lips and tongue thrust emerged. These involuntary movements at times recede for several weeks and remerge with undefined triggers. Her family history is significant for maternal and paternal generalized anxiety disorder and obsessive-compulsive disorder. Two maternal cousins display motor tics. Abigail does not present with depressive symptoms and disruptive behaviors. However, she displays a cautious and anxious temperament and obsessive thoughts.

Abigail's presentation meets the *Diagnostic and Statistical Manual of Mental Disorders, fifth edition* (DSM-5; American Psychiatric Association [APA], 2013) diagnostic criteria for persistent (chronic) motor tic disorder. Specifically, Abigail exhibits multiple motor tics that wax and wane. These have persisted for more than 1 year. Abigail also displays anxiety and obsessive thoughts, often comorbid with tic disorders.

Richard

Richard is 14 years old. His medical history and early development were unremarkable. At eight, he began to experience involuntary hand and knee movements. Now he has a wide range of vocal and motor tics that include sniffing, whistling, swearing, stretching his neck, rolling his hands, and stretching his legs. Richard has average intellectual and neuropsychological abilities with the exception of hyperactively and impulsivity as well as inattention. He has obsessive thoughts and compulsive behaviors as well as significant symptoms of anxiety, particularly related to concerns about humiliation, rejection, and social anxiety.

Richard's presentation meets DSM-5 diagnostic criteria for Tourette's disorder, displaying multiple motor and vocal tics that are both simple and complex. These tics have occurred for several years and are not better explained by another medical or condition or substances. Like many adolescents with Tourette's disorder, he has comorbid symptoms of attention-deficit/hyperactivity disorder and generalized anxiety disorder.

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Overview

Both Abigail and Richard demonstrate tics – "sudden, rapid, recurrent, nonrhythmic motor movements or vocalizations" (APA, 2013 p. 82). Tics often vary over time, waxing or waning, but at any point, the tic repertoire recurs in a characteristic fashion. Motor and vocal tics are classified as *simple* or *complex* (APA, 2013). Simple tics are short in duration, often milliseconds, whereas complex tics are longer in duration, lasting seconds, and may include a combination of simple tics. Simple motor tics may include jerking of the head, shoulder shrugging, eye blinking, or grimacing. Simple vocal tics may include grunting, barking, throat clearing, or sniffing. Complex motor tics often appear purposeful and can include jumping, stomping feet, *copropraxia* (sexual or obscene gesture), or *echopraxia* (imitation of someone else's movements). Similarly, complex vocal tics can present as *palilalia* (repeating one's own sounds), *echolalia* (repeating the last-heard word or phrase), or *coprolalia* (uttering social inappropriate words such as obscenities or ethnic, racial, or religious slurs) (see Table 21.1). Transient or fleeting tics are common during childhood, affecting approximately 11–20% of children (Cubo et al., 2011; Linazasoro, Van Blercom, & Ortiz De Zárate, 2006; Snider et al., 2002).

Typically, children initially display tic behaviors between 5 and 6 years of age, with peak severity around 10–12 years (Pappert, Goetz, Louis, Blasucci, & Leurgans, 2003; Peterson, Pine, Cohen, & Brook, 2001). The majority of children experience a decrease and/or remission of symptoms following puberty. Tic disorders also have a high comorbidity with attention-deficit/hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD), and other anxiety disorders, as well as behavioral or conduct problems (Centers for Disease Control and Prevention [CDC], 2009). One of three DSM-5-defined tic disorders, Tourette's disorder (TD), presents in approximately 3–8 per 1000 children (Scahill, Specht, & Page, 2014).

This chapter includes four sections: first, a review the history of Tourette's and tic disorders with a focus on the *Diagnostic and Statistical Manual of Mental Disorders* (DSM) diagnostic criteria; second, a discussion of research on prevalence rate, symptom presentation and developmental course, and etiology and mechanisms; third, a description of comorbid disorders that often present alongside tic disorders and functional impairments; and fourth, Tourette's disorder and tic disorder diagnostics and current behavioral, pharmacological, and other medical treatments.

History of Diagnosis

The earliest reported case of Tourette's disorder, also referred to as Tourette's syndrome, was in 1825 by Jean-Marc Itard. Itard described a French noblewoman who began to display motor tics at 7 years old (Hyde & Weinberger, 1995). Later in life, the young woman developed involuntary vocations of screams and strange cries, which

Table 21.1 Types of tics

	Vocal	Motor
Simple tics	Throat clearing Grunting Barking Sniffing/snorting	Head jerking Shoulder shrugging Blinking Grimacing
Complex tics	*Palilalia*, repeating one's own sounds *Echolalia*, repeating the last-heard word or phrase *Coprolalia*, uttering socially inappropriate words (e.g., obscenities or ethnic, racial, or religious slurs)	Combination of simple motor tics Jumping/stomping *Copropraxia*, sexual or obscene gestures *Echopraxia*, imitation of another's movements

eventually manifested into coprolalia (utterances of socially inappropriate words). Due to her behaviors, she lived a solitary life.

Fifty years after Itard's first description of motor tics, Jean-Martin Charcot, a French physician, and his resident Gilles de la Tourette presented a series of seven cases that outlined similar histories and symptoms, now associated with Tourette's disorder. These early descriptions by Charcot included the waxing and waning of multiple motor and vocal tics, an early age of onset, and the genetic influences of the disorder (Keen-Kim & Freimer, 2006).

Early understanding and treatment of Tourette's disorder was largely misunderstood. In the early 1940s, several case studies were published that conceptualized Tourette's disorder using a psychodynamic interpretation (e.g., Ascher, 1948; Mahler & Rangell, 1943). At that time, Tourette's disorder was considered a psychiatric disorder, as it seemed that patients were able to voluntarily suppress symptoms for a period of time. Because symptoms appeared to exacerbate during periods of stress and anxiety, symptom etiology was generally associated with psychological distress.

Tourette's disorder was first included in the third edition of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-III; APA, 1980). It was found in the chapter Disorders Usually First Evident in Infancy, Childhood, or Adolescence. Tic disorders comprised of Tourette's disorder, chronic motor tic disorder, transient tic disorder, and atypical tic disorder. Diagnostic features of all three disorders were the presence of recurrent, involuntary, repetitive, rapid, purposeless, motor movements and the ability to suppress movements voluntarily for minutes to hours. The DSM-III diagnostic differences included:

- *Tourette's disorder*: onset between 2 and 13 years of age, the presence of multiple vocal and motor tics, and duration over 1 year
- *Chronic motor tic disorder*: onset either in childhood or after 40 years of age, unvarying intensity of tics over weeks or months, and duration of at least 1 year
- *Transient tic disorder*: onset during childhood or adolescence and duration of tics of at least 1 month, but not more than 1 year
- *Atypical tic disorder*: used when the diagnoses of a tic disorder could not be adequately classified

Additional associated features of tic disorders included sham and self-consciousness. Furthermore, social and occupational impairment could be present owing to social ostracism and functional impairments due to tic movements. Revision of the third edition of the DSM (DSM-III-R; APA, 1987) included age of onset prior to age 21 as well as the elimination of substance abuse or medical condition as a causal factor.

In the fourth edition of the DSM (DSM-IV; APA, 1994), the definition of a tic remained unchanged. Diagnostic differences included (1) onset of tics prior to age 18, (2) the disturbance caused marked distressed or significant functional or social impairment, and (3) the individual never met criteria for a more severe tic disorder. Disorder diagnostics included:

- *Tourette's disorder*: multiple motor and one or more vocal tics that occur multiple times a day (usually in bouts) nearly every day or intermittently for a period of 1 year, without a tic-free duration of more than three consecutive months
- *Chronic motor or vocal tic disorder*: single or multiple motor or vocal tics, but not both, and duration of at least 1 year without a tic-free period of more than three consecutive months
- *Transient tic disorder*: single or multiple motor and/or vocal tics that occur multiple times a day, nearly every day, for at least 1 month and less than 1 year. Additionally, this disorder should be specified if there is a single or multiple episodes.

Tic disorder, not otherwise specified replaced *Atypical tic disorder*; however, the diagnostic criteria remained the same. The DSM-IV additionally outlined differences between simple and complex tics (see Table 21.1). In the text revision of the DSM-IV (DSM-IV-TR; APA, 2000), diagnostic criteria for tic disorders remained unchanged.

Current DSM-5 Definition

Tic disorders as defined by the fifth edition of the DSM (DSM-5; APA, 2013) include five diagnostic categories and have remained relatively similar to the previous editions. The disorders include, in hierarchical order of severity, Tourette's disorder, persistent (chronic) motor or vocal tic disorder, provisional tic disorder, and other specified and unspecified tic disorders. The diagnoses for Tourette's disorder, persistent (chronic) motor or vocal tic disorder, and provisional tic disorder are based on the following four criteria: Criterion A, the presence of motor and/or vocal tics; Criterion B, duration of symptoms; Criterion C, age of onset must be less than 18 years old; and Criterion D, absence of other known causes (e.g., medical, substance use). Criterion E specifies that a more severe tic disorder has never been met. Diagnostic differences between these three disorders include:

- *Tourette's disorder*: the presence of both motor and vocal tics for more than 1 year
- *Persistent* (*chronic*) *motor or vocal tic disorder*: single or multiple motor or vocal tics for more than 1 year, but not both motor and vocal
- *Provisional tic disorder*: single or multiple motor and/or vocal tics for less than 1 year

Lastly, criterion for *other specified* and *unspecified tic disorder* specifies that significant distress or functional impairment must not be present and the individual does not meet full diagnostic criteria for other tic disorders. For complete outline of diagnostic criteria for each tic disorder, see Table 21.2.

Table 21.2 Diagnostic criteria of tic disorders

Tourette's disorder	Criterion A: Both motor and one or more vocal tics have been present at some during the illness, although not necessarily concurrently Criterion B: Tics may vary in frequency but have persisted for greater than 1 year since first onset Criterion C: Onset prior to age 18 years Criterion D: The disturbance is not attributable to the effects of a substance or another medical condition
Persistent (chronic) motor or vocal tic disorder	Criterion A: Single or multiple motor tics or vocal tics have been present but not both motor and vocal Criterion B: Tics may vary in frequency but have persisted for greater than 1 year since first onset Criterion C: Onset prior to age 18 years Criterion D: The disturbance is not attributable to the effects of a substance or another medical condition Criterion E: Never met criteria for Tourette's disorder *Specify if* with motor tics only or with vocal tics only
Provisional tic disorder	Criterion A: Single or multiple motor and/or vocal tics Criterion B: Tics have persisted for less than 1 year since first onset Criterion C: Onset prior to age 18 years Criterion D: The disturbance is not attributable to the effects of a substance or another medical condition Criterion E: Never met criteria for Tourette's disorder or persistent (chronic) motor or vocal tic disorder
Other specified tic disorders	Criterion specifies that significant clinical distress or functional impairment must be present, but does not meet full diagnostic criteria for the other tic disorders or any of the disorders in the neurodevelopmental disorders diagnostic class. A situation for this specification is when clinicians choose to communicate reasons, the presentation does not meet full diagnostic criteria for a tic disorder
Unspecified tic disorders	Criterion specifies that significant clinical distress or functional impairment must be present, but does not meet full diagnostic criteria for the other tic disorders or any of the disorders in the neurodevelopmental disorders diagnostic class. A situation for this specification is when clinicians do not choose to communicate reasons, the presentation does not meet full diagnostic criteria for a tic disorder

Adapted from the DSM-5 (APA, 2013)

Review of Research

Prevalence

Transient tics (i.e., provisional tic disorder) are common in childhood, affecting approximately 11% to 20% of children (Cubo et al., 2011; Linazasoro et al., 2006; Snider et al., 2002). The male-to-female ratio in these studies was between 2:1 and 3.5:1. Due to the nature of cross-sectional studies, the number of children that later developed Tourette's disorder or a chronic tic disorder is unknown.

Community surveys of children provide estimates of Tourette's disorder between 0.5 and 38 cases per 1000 (CDC, 2009; Hirtz et al., 2007; Robertson, 2008; Scahill, Sukhodolsky, Williams, & Leckman, 2005). Scahill et al. (2014) noted the large variance in such surveys is likely a result of sampling methods, sample sizes, rate of subject participation, assessment methods, and definition criterion. Scahill et al. estimate a prevalence rate of 6 cases per 1000. Estimates of persistent (chronic) motor tic disorder range from 3 to 8 cases per 1000, whereas persistent (chronic) vocal tic disorder may be as high as 8 cases per 10,000 (Khalifa & Knorring, 2007; Kraft et al., 2012; Kurlan et al., 2001; Scahill, Williams, Schwab-Stone, Applegate, & Leckman, 2006).

Symptom Presentation/Developmental Course

Tic disorders generally first appear between 5 and 6 years of age. Symptoms tend to increase with severity peaking around 10–12 years of age, gradually decreasing following puberty and remising in late adolescence and adulthood (Pappert et al., 2003; Peterson et al., 2001). For those who continue to display symptoms as an adult, the severity is often mild. However, a small group of adults continue to display severe symptoms.

Tourette's disorder is the most severe of tic disorders. Symptom onset is often gradual, initially with simple motor tics, and progresses in a rostrocaudal direction (i.e., from the head down). Simple vocal tics tend to present 1 to 2 years after motor tic onset (Leckman, 2002). Motor and vocal tics increase in severity and complexity with age; however, symptoms can wax and wane (Peterson & Leckman, 1998). Leckman et al. (1998) found in adults diagnosed with Tourette's disorder as a child, one third experienced complete remission, less than half experienced minimal to mild tics, and less than one quarter experienced moderate or greater tics. Less than five percent of adults in Lekman's report experienced a worsening of tic symptoms as an adult when compared to their childhood.

By 10 years of age, most individuals with Tourette's disorder experience premonitory urges prior to a tic as well as sense of release following the tic (Leckman, 2002). Most individuals are able to temporarily suppress tics; however, this can detract from concentration and be exhausting (Himle, Woods, Conelea, Bauer, & Rice, 2007). The ability to suppress tics tends to increase with age. During periods of excitement, anxiety, stress, and fatigue, tic symptoms often exacerbate, whereas periods of focused concentration and fine motor movement (e.g., playing instruments, dancing, playing sports) decrease tic frequency (Conelea & Woods, 2008).

It is frequently reported that individuals with tic disorders also experience comorbid psychiatric disorders. These include attention-deficit/hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD), anxiety, depression, learning disabilities, and oppositional defiant and conduct disorder. Often these comorbid disorders result in increased impairment and may require psychotherapeutic or pharmacological intervention.

Etiology

Over the past four decades, Tourette's and tic disorders have become understood to have an organic etiology. Frequency of tics may be exacerbated by psychological distress (Conelea & Woods, 2008). Evidence from neuroimaging has mounted support that Tourette's disorder should be classified as a neurological movement disorder (Peterson et al., 2001). This supposition is

generated by the discovery of several biological markers that are linked to Tourette's disorder (Dietrich et al., 2015), as well as research suggesting abnormal brain morphology in those diagnosed with Tourette's disorder (e.g., Church & Schlaggar, 2014; Draganski et al., 2010; Osmon & Smerz, 2005; Tobe et al., 2010).

Family Studies

In a review of family studies, O'Rourke, Scharf, Yu, and Pauls (2009) found rates of Tourette's disorder in first-degree relatives ranging from 10 to 100 times higher than in the general population. Furthermore, rates of persistent (chronic) motor or vocal tic disorder range from 7% to 22% among first-degree relatives (O'Rourke et al., 2009). These high rates support that tic disorders are in part genetically related. Family studies have also demonstrated higher rates than population prevalence of first-degree family members of those with tic disorders additionally diagnosed with OCD and ADHD (O'Rourke et al., 2011).

Twin and Genetic Studies

Twin studies report monozygotic twin concordance range between 50% and 70%, whereas dizygotic twins range from 10% to 23% (Price, 1985). These findings are also supported by later research (Bolton, Rijsdijk, O'Connor, Perrin, & Eley, 2007; Hyde, Aaronson, Randolph, Rickler, & Weinberger, 1992). Genetic testing and the identification of specific genetic markers further support tic disorders as heritable. A full review of the literature on genetics of tic disorders is beyond the scope of this chapter and better described elsewhere (see: Dietrich et al., 2015; Pauls, Fernandez, Mathews, State, & Scharf, 2014).

Anatomical

Anatomical studies of those with Tourette's disorder are inconsistent due in part to the high comorbidity of psychiatric disorders. However, a growing number of studies suggest functional or structural abnormalities in frontal or striatal brain regions as well as the frontal-striatal relationships may be contributing factors (Osmon & Smerz, 2005). It is also surmised that malfunction of the cortico-striatal-thalamic-cortical loop contributes to Tourette's disorder (Jankovic, 2001).

Studies describe a decreased volume of the caudate nucleus in children with Tourette's disorder (Bloch, Leckman, Zhu, & Peterson, 2005; Hyde et al., 1995; Makki, Behen, Bhatt, Wilson, & Chugani, 2008; Peterson et al., 2003). Other subcortical studies have less consistent findings (Church & Schlaggar, 2014; Peterson et al., 1993, 2003). Church et al. (2014) suggest that the lack of consistent findings is suggestive of a communication problem between multiple nuclei within a circuit. Two studies report on cerebellum anatomical differences. Cerebellum volume analysis indicates no difference in the size in children or adults with tic disorder when compared to same-age normals (Hong et al., 2002; Tobe et al., 2010). However, Tobe et al. (2010) describe a gray matter reduction in the lateral cerebellar hemispheres. Cortical studies appear to have greater consistency. Largely, these studies provide evidence of cortical thinning or volume changes in the motor and frontal and parietal cortex in children, which persist into adulthood (Draganski et al., 2010; Fahim et al., 2010; Sowell et al., 2008). Diffusion tensor imaging studies of individuals with Tourette's disorder show differences around the corpus callosum (Cavanna et al., 2010; Draganski et al., 2010; Jackson et al., 2011; Liu et al., 2013), somatosensory and motor cortex (Cheng et al., 2014; Draganski et al., 2010; Thomalla et al., 2009), and caudate nucleus (Saporta et al., 2010).

Assessment

Tics may wax and wane in frequency making observation of tics difficult for professionals. Therefore, collecting information using multiple data sources, both objective and subjective, across a variety of settings and from multiple sources is highly beneficial. Observation records and video recording outside of a clinic setting by adults interacting with children who experience

tics can assist in a professional assessment of tic somatology and severity. Similarly, clinical observations and documentation of tics contribute to assessment of the disorder. Together, these data provide robust information to inform clinical decision-making.

Clinician-observer rating scales are used to assess various behaviors and symptoms associated with tics – tic type, duration, frequency, complexity, location, suppression, and impairment. Rating scales are completed by experienced and trained clinicians following a clinical interview. The most commonly used clinician-observer scales include the Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989), the Shapiro Tourette's Syndrome Severity Scale (STSSS; Shaprio & Shaprio, 1984), the Hopkins Motor and Vocal Tic Scale (HMVTS; Walkup, Rosenberg, Brown, & Singer, 1992), and the Tourette Syndrome Clinical Global Impression (TS-CGI; Leckman et al., 1989).

Self-report and parent-rating scales provide a subjective option in tic symptom presentation. Parent reports also assists in the evaluation of child's behaviors at home and in the community. Self-report scales include the Motor Tic, Obsessions and Compulsions, Vocal Tic Evaluation Survey (MOVES; Gaffney, Sieg, & Hellings, 1994) and the Premonitory Urge for Tics Scale (PUTS; Woods, Piacentini, Himle, & Chang, 2005). Parent-rating scales include the Parent Tic Questionnaire (PTQ; Chang, Himle, Tucker, Woods, & Piacentini, 2009), Child Tourette's Syndrome Impairment Scale: Parent Report about Child (CTIM-P; Storch 2007), Yale Tourette Syndrome Symptom List (TSSL; Cohen, 1980), and Tourette's Disorder Scale – Parent Rated (TODS-PR; Shytle et al., 2003).

The use of video-based rating scales provides objective data of a child's tics. Video records may include various tasks (e.g., reading, sitting alone, performing mathematics, placed under stress) performed in clinical and a variety of nonclinical settings. The Modified Rush Video-Based Tic Rating Scale (RVTRS; Goetz, Pappert, Louis, Raman, & Leurgans, 1999) is shown to result in effective clinical decision-making results.

Comorbid Disorders

Tic disorders are highly comorbid with ADHD and OCD. Less so, but also present with tic disorders are developmental disorders, disruptive behaviors, depression, anxiety, and learning disorders (APA, 2013). Of children surveyed in the United States by the Center for Disease Control (CDC; 2009), 79% were also diagnosed with at least one comorbid psychiatric or neurodevelopmental disorder. Specifically, 64% were also diagnosed with ADHD, 43% with behavior or conduct problems, 40% with anxiety, 36% with depression, and 28% with a developmental delay. Similarly, Roessner et al. (2007) found in a sample from 65 sites in 22 countries a comorbid rate of 61% for tic disorder with ADHD whereas a 19% rate for tic disorder with OCD, which are comparable to reports from Mol Debes, Hjalgrim, and Skov (2008) and Sukhodolsky et al. (2003). Roessner et al. (2007) report a higher comorbid rate of tic disorder with OCD in children (5–10 years of age) when compared to adolescents (11–17 years of age); comorbid mood disorders were found in greater frequency when ADHD coexisted. Specifically, comorbid tic disorders and conduct or oppositional defiant disorders had a greater prevalence with the coexistence of ADHD (Roessner et al., 2007). Rates of conduct disorder and oppositional defiant disorder increased year wise for this group as well.

Freeman and Tourette Syndrome International Database Consortium (2007) report that in those diagnosed with a comorbid tic disorder and ADHD, 7% met the predominantly hyperactive/impulsive presentation, 37% met the predominantly inattentive presentation, and 51% met combined presentation. In this group, those with a comorbid tic disorder-ADHD combined presentation or predominantly hyperactive/impulsive presentation demonstrated increased rates of problems with anger control. Furthermore, those with a comorbid tic disorder and ADHD had significantly greater social skill deficits as well as more frequent sleep problems than those without a diagnosis of ADHD.

The high comorbid rates of tic disorders with ADHD and OCD with tic disorders are largely believed to be attributed to neurological similarities. The high comorbidity rates of tic disorders with ADHD and with OCD are in part believed to result from similarities in cortico-striatal-thalamic-cortical dysfunction (Jankovic, 2001). Others purport genetic disposition impacts these comorbid rates (Eysturoy, Skov, & Debes, 2015).

Associated Impairments

Functional impairment is the inability to perform activities that are required for an individual's daily living. Tics may interfere with and therefore be the stimulus for or cause of functional impairment. Storch et al. (2007) found in a sample of 59 children (mean age = 11.4 years) parents report the highest functional impairment related to tics occurred in school (e.g., during class, doing homework, concentrating of work, and being prepared for class), followed by social activities. Impairment in social activities is primarily attributed to being teased by peers and difficulty making new friends. Further analysis found that close to 70% of problems that caused functional impairment experienced by this group were related to non-tic-related factors, specifically those associated with ADHD or OCD symptoms. Similarly, Bernard et al. (2009) found in a sample of 56 children ages 5–17 years old (mean age = 11 years) whose tic symptoms ranged from mild to severe that motor and vocal tics were not correlated to quality of life. However, ADHD and OCD symptoms were both significant contributors to quality of life. Debes et al. (2010) found children with Tourette's disorder comorbid with ADHD or OCD tend to have increased psychosocial and educational impairment. These studies suggest that for the majority of individuals with tic disorders, symptoms related to tics do not result in significant functional impairment, rather the comorbid disorders result in greater impairment than the tics themselves.

Individuals with tic disorders do not generally exhibit impairment in general intellectual ability (Como, 2001). The majority of children with tic disorders demonstrate neuropsychological functioning within normal limits (Yeates & Bornstein, 1996). However, for those who do exhibit impaired neuropsychological functioning, it is difficult to determine if the impairment is a result of a comorbid disorder (e.g., ADHD, OCD, mood disorder, developmental disorder) or from a fronto-striatal dysfunction, associated with tic disorders (Osmon & Smerz, 2005). Neuropsychological impairments that are most commonly associated with tic disorders are impairment in fine motor skills (Como, 2001) and speed of executing movement (Georgiou, Bradshaw, Phillips, Bradshaw, & Chiu, 1995).

Differential Diagnosis

The DSM-5 outlines four primary diagnostic considerations that should be evaluated for a differential diagnosis (APA, 2013). The first consideration is abnormal movement that may accompany other medical conditions and stereotypic movement disorder, defined as "involuntary rhythmic, repetitive, predictable movements that appears purposeful but serve no obvious adaptive function or purpose and stop with distraction" (APA, 2013 p. 84). They are not preceded by a premonitory urge. Examples of stereotypies include hand flapping or rotating, arm waving, finger wiggling, rocking, and head nodding. Chorea presents as bilateral movements affecting most of the body and are rapid, random, continual abrupt, irregular, unpredictable, non-stereotyped actions. These movements vary in timing, direction, and distribution of movements. Lastly, dystonia is defined as involuntary muscle contractions that result in slow respective movements or abnormal postures.

The second diagnostic consideration includes substance-induced and paroxysmal dyskinesias (APA, 2013). Paroxysmal dyskinesias are a group of rare hyperkinetic movement disorders that are characterized by their episodic nature (Unterberger & Trinka, 2008). They are sudden involuntary abnormal movements that often arise from normal motor actively. Duration of events can last from seconds to several hours.

The third diagnostic consideration is myoclonus, which is the sudden unidirectional movement that is nonrhythmic (APA, 2013). Myoclonic twitches often occur due to sudden muscle contractions or reaction. They often present alone or in a sequence that can last a few minutes and cannot be controlled.

Lastly, OCD and related disorders need to be ruled out (APA, 2013). Specifically, the difference between OCD and tic disorders is outlined in the DSM-5. OCD has a cognitive drive to perform an action, whereas tic disorders do not possess this drive, despite the premonitory urge. In addition, impulse-control problems and other repetitive behaviors, such as nail biting, skin picking, and hair pulling, are goal driven and more complex in movement than tics.

Treatment

The primary treatment for the majority of individuals with tic disorders is behavioral therapy. The therapeutic modalities that have developed the strongest support for tic disorders are habit reversal training (HRT) and its expanded version, comprehensive behavioral therapy (CBIT) for tics, as well as exposer with response prevention (ERP). Some individuals may require pharmacological intervention. The use of deep brain stimulation (DBS) is prescribed for those with severe symptoms.

The goal of HRT (Azrin & Nunn, 1973) is to teach the patient to recognize the occurrence of each tic and learn pre-tic warning signs and then engage in a response that interrupts the tic. For example, a child who experiences a sniffing tic learns to identify the premonitory urge, and in response to this warning, he learns to exhale as a means to inhibit the tic. HRT has three primary components consisting of awareness training, competing response training, and social support. HRT is typically conducted over eight to 11 therapy sessions targeted at one tic per week and the practice exercises between sessions. Numerous studies demonstrate HRT effectiveness (e.g., Bate, Malouff, Thorsteinsson, & Bhullar, 2011; Cook & Blacher, 2007; Piacentini et al., 2010; Wilhelm et al., 2012; Woods, Conelea, & Himle, 2010).

The second therapeutic model is comprehensive behavioral therapy for tics (CBIT) and is expanded from the HRT model (Woods, 2008). This is a manualized treatment that utilizes an individualized function-based assessment to evaluate contextual factors that may exacerbate tics. Modifications are introduced to eliminate or alter the factors that exacerbate tics. The modifications may be coupled with traditional HRT strategies. Relaxation skills are also thought to assist with decreasing anxiety. For young children, CBIT is combined with behavioral reward systems to facilitate participation. Similar to HRT, CBIT is found to be effective in treating tic disorders (Piacentini et al., 2010; Scahill et al., 2013; Wilhelm et al., 2012; Woods et al., 2011).

Exposure and response prevention (ERP) is a behavioral therapy for obsessive-compulsive disorder shown effective in treating tics (Cook & Blacher, 2007; Verdellen, Keijsers, Cath, & Hoogduin, 2004). ERP exposes patients to anxiety-provoking stimuli that can exacerbate tics and teaches the patient to repress a tic for an extended period of time (e.g., 2 h). The effect differs from suppressing tics for short durations (30 s). The rationale behind ERP is to simultaneously teach the patient to reframe from releasing tics, which will allow the patient to habituate the pre-tic sensation and then suppress the tic response through repeated practice and prolonged tic suppression.

For the majority of individuals with tic disorders, pharmacological treatment is not warranted. Cheung et al. (2007) suggest the following considerations prior to pharmacological intervention: (1) social impairment, (2) functional impairment, (3) pain, and (4) neurological or harmful behaviors, secondary to the tics (e.g., self-injurious behavior). The first-tier medications often include guanfacine and clonidine (Gilbert & Jankovic, 2014). However, for some, these medications can increase tic severity and should be monitored closely. A full review of medications for treating tic disorders is beyond

the scope of this chapter and is described elsewhere (see: Karen, Jonathan, Cristiano, Umberto, & Eugenio, 2013; Roessner et al., 2007; Weisman, Qureshi, Leckman, Scahill, & Bloch, 2013).

Introduced in 1999, deep brain stimulation (DBS) has been used as a treatment for those with tics who have a medication and behavioral therapy refractory response (Vandewalle, van der Linden, Groenewegen, & Caemaert, 1999). DBS is a surgical intervention, and there are specific recommendations for pre- and postoperative evaluations to be conducted by a multidisciplinary team (see: Schrock et al., 2015). Although the number of patients who have underwent DBS is limited, results show promise for this treatment approach (Visser-Vandewalle et al., 2014).

Lastly, due to the high comorbid rate of tic disorders and other psychiatric disorders, psychotherapeutic and psychopharmacological treatments are highly recommended to address symptoms associated with the comorbid disorders. This is especially important as these comorbid disorders often result in higher levels of impairment compared to tics alone. Research is yet to demonstrate differing treatment recommendations for the comorbid disorder for those with and without tics.

Prognosis

The prognosis for children with tic disorders is promising in that most children experience a decrease or complete remission of tic symptoms during adolescence and early adulthood. It is rare to observe an a consistent increase in tic symptoms across a lifespan. Current research describes behavioral therapies that result in decreasing or managing tic symptoms. Tics themselves rarely result in impairment. It is important however to understand that tics may induce social impairments when peers do not understand the reason for tic behaviors which can result in teasing or bulling. For individuals with tic disorders and comorbid psychiatric disorders, these comorbid disorders frequently require additional interventions.

Abigail and Richard

Abigail, from the beginning of the chapter, is 7 years old and began displaying motor tics at 5 years old. She also displays anxiety and obsessive thoughts. She will likely require behavioral therapy to decrease tics. However, she will also likely require intervention to assist in development of more effective coping skills to manage anxiety and obsessive thoughts. Despite her early display of tics, her overall prognosis is good as intervention can be provided early for her tics, which have not increased in severity, as well as to decrease her anxiety and obsessive thoughts.

Richard, the 14-year-old with increasing simple and complex motor and vocal tics, will likely require more intensive intervention. Despite hitting puberty, where most youth experience a decrease in symptoms, he continues to display an increase in tic severity and frequency. His tics are coupled with symptoms of ADHD and OCD. He experience significant anxiety related to humiliation, rejection, and social anxiety. He will likely require behavioral therapy targeted at decreasing tic symptoms as well as psychotherapy targeting comorbid disorders. Given his severity, Richard may require a trial of medication aimed at reducing tic symptoms. Richard's prognosis is not as positive as Abigail's giving the severity of tic symptoms and presence of comorbid disorders. Richard will likely continue to experience some impairment from his symptoms throughout his adolescent and perhaps into adulthood.

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Developmental Coordination Disorder

22

Cheryl Missiuna, Lisa Rivard, and Wenonah Campbell

What Is Developmental Coordination Disorder?

"There is something wrong with my child". What physician hasn't heard these words at some point from a parent whose preschool or school-aged child seems to be developing well, may have achieved major motor and cognitive milestones on time … but where something is just not right? The concerns begin when parents notice their toddler sitting back and watching, while other children climb on play equipment or manipulate toys. Instead of supervising from a nearby bench, parents find themselves standing at the bottom of the ladder of the slide, ready to catch their child when he falls off. These same parents are still helping to put coats and boots on when the child is six, tying shoelaces at eight, cutting meat at ten and still encouraging pants with an elastic waist at age twelve instead of jeans. It is puzzling. These children are often of average or above-average intellectual ability, are well supported and stimulated in their home environment, walked on schedule, give good eye contact, interact well and look quite normal physically. Parents usually raise concerns with their primary health-care provider before their child enters school, but the issues are often vague (Missiuna, Moll, Law, King, & King, 2006). "He's tired a lot", "She gets frustrated easily", "He doesn't want to go out and play", "Should she still be having toileting accidents in Grade 2?", "She whines, and wants me to do everything for her", "He falls all the time, and has lots of cuts and bruises". These children usually achieve basic motor milestones like sitting or standing on time but are slow to learn new motor skills such as holding and using a spoon, fork, crayon, pencil, pair of scissors; doing up buttons, zippers, shoelaces; learning to go downstairs with alternating feet, to ride a bicycle, to turn a doorknob ... everyday motor skills that require coordination (Missiuna, Moll, King, King, & Law, 2006). Children with these motor challenges have developmental coordination disorder (DCD).

Overview

DCD is a neurodevelopmental disorder that affects five to six percent of all school-aged children, with approximately two percent being severely impacted by their coordination challenges (Lingam, Hunt, Golding, Jongmans, & Emond, 2009). Children with DCD have difficulty mastering motor skills such as kicking a ball, or zipping a knapsack, and are unable to perform age-appropriate academic, leisure and

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self-care tasks, even when they have had many opportunities to learn and practice. Frequently described as "clumsy" or "awkward" by parents and teachers, these children tend to avoid physical activity (Cairney, Hay, Veldhuizen, Missiuna, & Faught, 2010), may show evidence of decreased strength and endurance over time (Raynor, 2001; Rivilis et al., 2011), and are at increased risk of becoming overweight (Cairney, Hay, Veldhuizen, Mahlberg et al., 2010). Even though children with DCD usually have typical or above-average intellectual abilities, their academic progress, social integration and emotional development can be seriously limited by their motor difficulties and by the frustration they experience when they cannot execute and complete basic daily activities (Mandich, Polatajko, & Roger, 2003; Missiuna, Rivard, & Pollock, 2011). DCD is a permanent and multifaceted health condition that persists through adolescence into adulthood (Cousins & Smyth, 2003; Kirby, Edwards, & Sugden, 2011; Kirby, Sugden, Beveridge, & Edwards, 2008; Rasmussen & Gillberg, 2000). By definition, children with DCD do not have any other identifiable medical or neurological condition that better explains their coordination problems. However, DCD is associated with other neurodevelopmental conditions, including ADHD, speech and language impairments (SLI) and learning disabilities (LD) (see "Associated Impairments and Comorbidities"). The persistence of DCD into early and later adulthood has been confirmed; in fact, adults with DCD may be denied many crucial life experiences such as driving a vehicle (Kirby et al., 2008). Although there is currently no cure for DCD, early intervention and management may help to reduce the serious emotional, physical and social consequences that often accompany this disorder.

History of the Diagnosis

Historically, Orton (1937) was the first to describe a syndrome of clumsiness as a common developmental disorder. Yet case studies and more systematic empirical work did not begin to appear in the literature until the 1960s. In 1975, Gubbay introduced the term "clumsy child syndrome" to describe a group of children whose motor coordination difficulties interfered with their ability to function at school but who had normal or above-average intelligence and no other medical conditions that would explain these difficulties. Since that time, children with motor coordination problems have been described using many labels including: developmental dyspraxia, minimal brain dysfunction, sensory integrative dysfunction, perceptuomotor dysfunction, physically awkward, and specific developmental disorder of motor function (Missiuna & Polatajko, 1995).

As health professionals began sharing research evidence across countries and disciplines, this wide variation in terminology led to confusion. Without a consistent label, it became difficult to establish diagnostic criteria. In 1987, the *Diagnostic and Statistical Manual of Mental Disorders* (3rd edition; *DSM–III*; American Psychiatric Association [APA], 1987) introduced the Motor Skills Disorder 315.40, "developmental coordination disorder", a term that was retained in the revised edition 2 years later (3rd edition revised; *DSM–III-R*; APA, 1989). The manual noted that the "essential feature of this disorder is a marked impairment in the development of motor coordination that is not explainable by Mental Retardation and that is not due to a known physical disorder" (pp. 48–49). Importantly, it stated that the diagnosis could only be made if the disorder significantly interfered with academic achievement or activities of daily living. Clumsiness alone was insufficient to warrant a diagnosis – there needed to be an impact on everyday function.

Despite its inclusion in DSM-III (APA, 1987), DCD did not really gain legitimacy as a chronic health condition that warranted diagnosis until 1994 when an international consensus meeting was held in London, Ontario, Canada. At that meeting, researchers and clinical experts from around the world made a decision that they would recognize all "clumsy" children as having developmental coordination disorder (DCD) and that they would use the term DCD to describe participants in their studies (Polatajko, Fox, & Missiuna, 1995). Not coincidentally, the meeting was held in the year that DSM-IV (4th edition; APA, 1994)

was released – an edition in which the term DCD and a description of the disorder was retained. By this time, diagnostic criteria for DCD had been expanded and developed. Instead of three criteria, there were now four. A delay in achieving motor milestones and/or the presence of clumsiness or motor impairment was still central to the diagnosis, as was the instruction to ensure that the motor impairment impacted on everyday activity. However, the physician was provided with more stringent "rule-out" criteria, including cerebral palsy, hemiplegia, and muscular dystrophy, and also pervasive developmental disorder. Of interest, instead of eliminating the diagnosis of DCD for all children who had an intelligence quotient below average, DSM-IV (APA, 1994) suggested that such children could be diagnosed with DCD, if their "motor difficulties were in excess of those usually associated with [intellectual delay]" (p. 54). The text revision to DSM-IV in 2000 did not change, and the four diagnostic criteria outlined became the gold standard for clinicians and researchers (Geuze, Jongmans, Schoemaker, & Smits-Engelsman, 2001). In 2001, Geuze and colleagues reviewed all studies which had applied these four diagnostic criteria for DCD when selecting research study participants and subsequently recommended that an intelligence quotient of 70 be established as the minimum for children participating in research studies. This recommendation was adopted and influenced participant selection and inclusion in research studies for the next two decades. Despite ongoing debate about parental preference for particular diagnostic terms (Miyahara & Register, 2000), use of the term DCD was again confirmed by an international group of researchers and clinicians at the Leeds Consensus in 2006 (Sugden, 2006). A systematic review by Magalhaes and colleagues in 2006 demonstrated that research articles about children with DCD had increased tenfold between 1995 and 2005 and that more than half now referred to children with this type of profile as children with DCD (Magalhaes, Missiuna, & Wong, 2006). It would seem that the term DCD was here to stay.

Given the length of time between editions of the DSM, and the accumulating evidence about DCD in the meantime, it is not surprising that leaders in the field decided to move ahead in providing guidelines on the definition, diagnosis and intervention of DCD (Blank, Smits-Engelsman, Polatjko, & Wilson, 2011). Recommendations were generated in 2010 at two consensus conferences in Germany and, following the endorsement of an international panel of experts, were adopted as the guidelines of the European Academy for Childhood Disability (EACD). The document provides 32 key recommendations arising from the evidence. While systematic and thorough, implementation of the guidelines for assessment and intervention requires extensive testing of the child and access to a multidisciplinary team of experts, which is unrealistic for many jurisdictions. However, a strength of the guidelines is their emphasis on considering comorbidities and on recommending evidence-based interventions and education of parents and teachers (Blank et al., 2011).

Finally, introduced in 2013, DSM-5 includes four new criteria for a diagnosis of DCD. Adapted from the Diagnostic Criteria 315.4 (F82) in DSM-5 (2013), these may be summarized as:

Criterion A: The acquisition and execution of coordinated motor skills is below those expected given the child's age and their opportunity for skill learning and use.

Criterion B: The motor difficulties significantly interfere with age-appropriate performance of activities of daily living (self-care) and impact on academic productivity, prevocational and vocational activities, leisure and play.

Criterion C: The onset is in the early developmental period.

Criterion D: The motor coordination difficulties are not better explained by intellectual delay, visual impairment, or other neurological conditions that affect movement.

DSM-5 (2013) reflects the first major change in diagnostic criteria for children with motor coordination difficulties since 1994. Criterion A emphasizes the ongoing difficulty that children with DCD have in acquiring and performing new motor skills despite having had experience and

opportunities for practice. This criterion implies that teaching of the new motor skill may be necessary; as we will see in later sections of this chapter, such teaching is not necessary for typically developing children when they learn new motor skills. The question of how much practice is needed before considering a DCD diagnosis presents a dilemma for practitioners but challenges them to think about the child's home environment and prior experiences.

Perhaps the most important change in the DSM-5 diagnostic criteria is the recognition that the motor coordination difficulties impact on more than self-care and academic activities, extending to the playground as well. No longer focusing solely on productivity in school, the impact of DCD on the home, the family and the child's ability to participate in community activities is clearly acknowledged. The fact that DCD is a chronic, life-long health condition is also reflected strongly in Criterion B.

Reflecting the many studies that have shown clear evidence of the presence of DCD in preschool children (Wilson et al., 2014), Criterion C now specifies that the onset of these motor coordination challenges may be seen in the early developmental period. This emphasizes the need for physicians to inquire about early observations and delays, not just in attainment of basic motor milestones such as sitting or standing, but in learning to explore furniture or play equipment, or in learning to manage spoons and forks, or to toilet and dress independently.

Finally, Criterion D challenges the physician to think about whether other possible explanations such as significant intellectual delay or visual impairment might better explain the observed motor coordination difficulties (see "Screening and Diagnosis" section). If more than one diagnosis is present (e.g. ADHD), physicians are encouraged to give a diagnosis of DCD in addition to other relevant diagnoses to ensure that motor coordination difficulties are recognized and managed as part of comprehensive treatment.

In summary, the DSM-5 diagnostic criteria recognize two decades of research demonstrating that DCD involves impairment in both the learning and ongoing execution of motor skills; the coordination difficulties impact all aspects of daily life at school, at home and in the community; DCD is a chronic condition that continues to impact on successful participation in activities throughout the lifespan; it can and should be recognized in the early years; and it is a developmental health condition in its own right, distinct from other causes of motor impairment.

Review of Research Evidence

Prevalence International research performed over the years has confirmed that large numbers of children are affected by DCD (Forsyth et al., 2007; Kadesjo & Gillberg, 1999; Lingam et al., 2009), with prevalence rates ranging from 1.4 to 15%. Lingam and colleagues, using DSM-IV criteria, proposed that 1.8% of school-aged children have severe DCD and 3% more probably have DCD (Lingam et al., 2009). Studies in other countries have found higher or lower numbers (reviewed in Forsyth et al., 2007), but the DSM has consistently suggested a mid-range estimate of prevalence among school-aged children of 5–6% (APA, 1994, 2000, 2013).

Although the motor skills that children are expected to learn differ on different continents, the country of origin does not have an impact on the prevalence of DCD. Previously it was thought that socioeconomic status did not have a bearing either; however, Lingam et al. (2009) reported a greater risk of DCD among children from rental housing and a lower maternal socioeconomic group, possibly reflecting a higher rate of preterm births or low birth weight. The number of children who present with motor coordination difficulties is much higher among children who are of low birthweight or born prematurely (Dewey et al., 2011; Edwards et al., 2011; Roberts et al., 2011; Williams, Lee, & Anderson, 2010). Depending upon the method of measurement, the prevalence of DCD can be influenced by obesity (Cairney, Hay, Veldhuizen, & Faught, 2011; Fong et al., 2011; Wagner et al., 2011; Zhu, Wu, & Cairney, 2011) as well as by the presence of co-occurring or comorbid developmental disorders. This is particularly true for DCD and ADHD. Missiuna and

colleagues found that 51% of children with ADHD had DCD (Missiuna, Cairney et al., 2014). A study by Pieters et al. (2012) illustrates that comorbidity of motor coordination difficulties with other neurodevelopmental disorders is not unusual and that developmental differences are often not specific to one domain. It has commonly been accepted that boys with DCD outnumber girls in a 2:1 ratio, as is often seen in other developmental disorders; yet, recent population-based studies of children with DCD have suggested that more equal numbers of boys and girls may be affected (Cairney et al., 2007; Missiuna, Cairney et al., 2011).

There has been a perception that prevalence rates may decline as children get older; however, there is no theoretical reason why the prevalence of DCD should change in adolescence or adulthood. Persistence of DCD into early and later adult life has been confirmed (Kirby et al., 2008; Kirby et al., 2011). The "perceived" decline in prevalence is more likely associated with the changing expectations that society places on children as they mature. Older children have more options for leisure or sports activities and may be more skilful at compensating for physical shortcomings (Missiuna, Moll, King, Stewart, & Macdonald, 2008). As a result, their coordination difficulties may impact their daily activities in a less noticeable way, giving rise to a false sense of improvement. In the past, this "improvement" was used as evidence that children would "grow out of" their motor coordination difficulties. Research would strongly encourage physicians not to tell parents that DCD will "go away".

Symptom Presentation Compared to their peers, children with DCD move more slowly with longer reaction times; they are inconsistent and inaccurate in their motor performance especially when movement demands are more complex and they have difficulty maintaining posture and balance (Astill & Utley, 2008; Johnston, Burns, Brauer, & Richardson, 2002; Jucaite, Fernell, Forssberg, & Hadders-Algra, 2003; Mackenzie et al., 2008; Mak, 2010; Rosengren et al., 2009; Williams, 2002). Children with DCD have been noted to activate different musculature and employ atypical muscle sequencing during motor performance, making coordination across multiple joints challenging (Johnston et al., 2002). To compensate, some children rigidly "fix" their joints, making them look awkward and stiff (the "Tin Man"). These children often trip or get hit by moving objects because they do not modify their body position quickly enough. Even with tasks where children only need to guide a single joint, children with DCD experience difficulties. A complex task such as handwriting, which requires children to control several joints at once (at the shoulder, elbow, wrist, fingers), is extremely difficult for them. Here you will often observe children "fixing" joints and holding their pencil very tightly. As they fixate joints to stabilize their bodies, their movements become inherently more rigid and jerky and less fluent (Astill, 2007; Astill & Utley, 2006; Missiuna, Rivard, & Bartlett, 2003).

In what seems to be a contradictory presentation, some children with DCD have low tone and actually look floppy; they seem to be constantly moving because they have trouble coordinating the activation of their postural muscles, especially their trunk (the "Scarecrow"). Children who have this latter problem may be described by teachers as "noodle" kids who fall off their chairs in class, are not able to sit up at circle time and frequently bump into other children in line. In either case ("Tin Man" or "Scarecrow"), their physical presentation looks awkward or clumsy because the coordination across multiple joints is not smooth or effortless. Overall, these less effective movements can cause them to fatigue more easily than children their age (Faught et al., 2013; Williams, 2002).

With a restricted set of movement abilities, children with DCD are less able to adapt to changes in their environment than their peers (Astill & Utley, 2006; Kagerer, Bo, Contreras-Vidal, & Clark, 2004). For example, they may have difficulty avoiding other people who are moving around them in close proximity; if the ground is uneven or a curb is higher than usual, they are more apt to trip. Difficulties in new motor learning mean that children with DCD are challenged when transferring motor skills that they have already learned to new situations with different movement parameters, or to a different environmental context (Bo & Lee, 2013;

Jelsma, Ferguson, Smits-Engelsman, & Geuse, 2015; Niemeijer, Smits-Engelsman, Reynders, & Schoemaker, 2003). Knowing how to catch a large ball does not mean that they can transfer those skills to catch a small ball; bouncing a ball in the gym may not generalize to the playground or to playing in a basketball game. Their inconsistent performance results, in part, from a tendency to repeat tasks without correcting errors in their performance; specifically, they fail to see task similarities and do not use feedback from previous actions to anticipate what they need to do on a subsequent occasion. In addition, they prefer to use vision over other forms of feedback (e.g. proprioceptive, kinesthetic), which means that they do not progress to more skilled stages of motor learning (Biancotto, Skabar, Bulgheroni, Carrozzi, & Zoia, 2011; Deconinck et al., 2006; van Waelvelde et al., 2006).

Etiology and Mechanisms Although much of the empirical research in DCD has been dedicated to investigating its underlying neural mechanisms, a definitive etiology and pathogenesis have yet to be determined (Visser, 2003; Zwicker, Missiuna, & Boyd, 2009). Children with DCD have deficits in many areas of performance including postural and gait control, visual-spatial processing, timing and motor planning and execution. Each of these difficulties has been suggested as a possible causal factor for DCD. However, comparisons of the findings across empirical studies investigating mechanisms have been problematic due to differences in study populations as well as the tasks used. Given the complex and heterogeneous nature of the disorder, especially the presence of co-occurring developmental conditions, it is unlikely that a single deficit will explain the motor and associated impairments that are observed in children with DCD. Speculations regarding the nature of the motor deficits in children with DCD have varied, depending upon the perspective taken and the specific processes examined. Over the years, several hypotheses have been proposed in the literature. These are summarized in Table 22.1.

Table 22.1 Proposed hypotheses regarding developmental coordination disorder (DCD) etiology and mechanisms

Hypothesis	Key points and supporting evidence
Delayed maturation	DCD was once believed to be delayed maturation in children who, with sufficient time and experience, would "catch up" to their well-coordinated peers. Multiple studies, including those with longitudinal study designs, have strongly disproved this idea (Losse et al., 1991; Rasmussen & Gillberg, 2000). Rather, the gap between the development of children with DCD and their peers appears to widen with age, particularly for severe DCD. Today it is recognized that DCD is a long-term problem that, if not identified early and managed appropriately, can inhibit the development of motor learning and increase the risk of secondary problems in other life spheres (see "Etiology and Mechanisms" section)
Praxis (motor planning) difficulty	Many years ago, Hodge (1998) proposed that difficulties with praxis, or motor planning, may be a shared factor in some children who present with motor and speech-language impairments. These researchers focused on a small subgroup of children with DCD who had co-occurring developmental apraxia of speech. These children are *not* representative of the majority of children with DCD and should be considered a distinct subset of the DCD population. Interestingly, researchers have recently revisited the concept of praxis. Gheysen, van Waelvelde and Fias (2011) have suggested that it is the motor planning stage that causes problems for children with DCD when trying to sequence a motor movement
Sensory-perceptual dysfunction	During the 1980s and 1990s, research adhering to an information-processing framework focused on sensory/perceptual impairment(s) as a possible etiology for DCD. These studies uncovered visual, proprioceptive, kinesthetic and cross-modal perception/integration deficits (Wilson & Mackenzie, 1998). Recent work has extended findings implicating a range of visual difficulties in DCD including problems with visual-spatial processing and memory, and a dependence on vision during motor performance (Alloway, 2007; Biancotto et al., 2011; Wilson, Ruddock, Smits-Engelsman, Polatajko, & Blank, 2013; Zoia, Castiello, Blason, & Scabar, 2005). Research into the role of vision continues today using more contemporary theoretical underpinnings and integrating more comprehensive accounts of motor control and learning (Rivard, Lee, Wishart, & Missiuna, 2017)

Hypothesis	Key points and supporting evidence
Atypical brain development	Several researchers have proposed that the comorbid presentation of many childhood developmental disorders, including DCD, is so common that there must be a shared factor (Gilger & Kaplan, 2001; Kaplan, Wilson, Dewey, & Crawford, 1998). They referred to this comorbid presentation as atypical brain development (ABD). In this model, speech-language, attentional and motor difficulties are grouped under one label and are considered variable expressions of one common factor. While this hypothesis may explain DCD subtypes and suggest why severely affected children are more likely to have comorbidities, it does not account for occurrences of DCD alone. Furthermore, this concept and terminology fell out of favour with efforts to encourage consistent use of the term DCD. However, researchers still discuss the ABD model today, given the consistent pattern of motor and associated impairments in many studies of children with DCD (Wilson et al., 2013)
Deficits in attention, motor control and perception (DAMP)	Similar to the ABD concept, the term "DAMP" (deficits in attention, motor control and perception) has been used to describe children with motor and other impairments (Gillberg, 2003; Pereira, Landgren, Gillberg, & Forssberg, 2001).This concept also recognizes the interplay between the cortical structures involved in attention, motor and perceptual functioning. As with ABD, DAMP conceptualizes comorbid difficulties as one generalized disorder. In Scandinavian countries, DAMP is still the preferred diagnostic label when motor and associated difficulties, particularly attention, are present
Automatization deficit	The automatization deficit hypothesis, initially proposed for children with dyslexia (Nicolson, Fawcett, & Dean, 2001), describes a difficulty in making motor tasks automatic when cognitive demands are increased (Tsai, Pan, Cherng, & Wu, 2009). According to this hypothesis, children with DCD must direct attentional resources to control the motor movements that are typically performed without conscious thought. The lack of automatization during motor performance becomes a challenge when a cognitive task is introduced, because the child with DCD does not have the attentional resources to devote to more than one task at a time (e.g. printing a spelling word is more difficult as the child with DCD must give conscious attention to how to hold the pen and form the letters).This hypothesis seems plausible in relation to DCD and has been validated in several research studies (Cherng, Liang, Chen, & Chen, 2009; Laufer, Ashkenazi, & Josman, 2008)
Internal modelling deficit	The internal modelling deficit hypothesis unites research using diverse tasks, experimental paradigms and theoretical frameworks (Adams, Lust, Wilson, & Steenburgen, 2014; Gabbard & Bobbio, 2011). Internal models are critical for predictive motor control and motor learning over time, comparing movement estimates with movement during performance to make real-time adjustments (Wilson et al., 2013). Across many motor tasks, children with DCD show poor predictive control and an inability to adapt to varying environmental conditions (e.g. changes to vision), suggesting that they do not update their internal models (Adams et al., 2014; Brookes, Nicolson, & Fawcett, 2007; Cantin, Polatajko, Thach, & Jaglal, 2007; Kagerer, Contreras-Vidal, Bo, & Clark, 2006). Research evidence continues to grow and will further refine our understanding of the use of internal models in DCD

Data from neuroimaging studies of children with DCD have recently become available and are contributing to knowledge of the neurobiology of DCD (Peters, Maathuis, & Hadders-Algra, 2013). Results of these studies suggest that the integration of messages from different neural structures is atypical in children with DCD. In one fMRI study, the level of brain activity in the parietal cortex was lower in children with DCD than in healthy children when completing a tracking task (Kashiwagi, Iwaki, Narumi, Tamai, & Suzuki, 2009). Another study required children with DCD and typically developing children to complete a fine motor tracing task under fMRI. A significantly greater level of brain activation was recorded, along with different brain areas, in children with DCD (Zwicker, Missiuna, Harris, & Boyd, 2010; Zwicker, Missiuna, Harris, & Boyd, 2011). Other researchers have found that the specialized functioning of the brain hemispheres, especially with respect to attention and the inhibition of motor responses, is atypical in children with DCD and may distinguish them from control children (Querne et al., 2008). A recent fMRI study of children with DCD, with and without co-occurring ADHD, suggests that

alterations in common neural pathways may underlie combined DCD and ADHD (McLeod, Langevin, Goodyear, & Dewey, 2014). Further neuroimaging work utilizing different motor tasks and cognitive demands will help to continue to advance research in this area and to delineate the potential cortical mechanisms involved in DCD.

Genetics Population-based information on whether or not DCD is inherited is still lacking. A familial component, or clustering, is thought to be involved in motor coordination problems, but little is known about the genetic factors or the relationship between genetic and environmental factors. A heritability estimate of 70% was obtained for DCD in one twin study, with monozygotic twins having higher concordance rates for DCD than dizygotic twins (Martin, Piek, Baynam, Levy, & Hay, 2010). Further twin studies using monozygotic designs have shown high levels of co-occurrence of DCD with anxiety (Pearsall-Jones et al., 2011), depressive symptomatology (Fliers et al., 2009; Piek et al., 2007) and a variety of other neuropsychiatric disorders, including autism (Lichtenstein, Carlström, Råstam, Gillberg, & Anckarsäter, 2010). Most studies of genetics have focused on children who have ADHD and have simply observed the frequent co-occurrence of DCD, without drawing any definitive conclusions (Deng et al., 2014; Fliers et al., 2009).

Developmental Progression: What Do Children with DCD Look Like Over Time?

In contrast to what early studies had suggested, children with DCD may achieve basic motor milestones on time (e.g. sitting, standing). Although a recent study by Lingham and colleagues (2009) suggested that late walking might be an early sign, in many children, walking is still achieved within normal limits. Children with DCD often first show significant motor difficulties when they are required to learn and perform complex skills. Skilled activities vary from one culture to another, but what they share in common is that these skills must be learned from caregivers or other children. At toddler or preschool age, difficulties can be seen as the child tries to manage utensils, carry or manipulate a toy, scribble with a crayon, or climb stairs with alternating feet. Self-care skills are consistently delayed. Children will have difficulty with tasks such as putting on and removing clothing and feeding themselves independently with utensils (they tend to use their fingers). By school age, children should have acquired basic motor skills, and it is expected that they will simply learn to apply them in new tasks and situations. This is the age when children with DCD begin to look noticeably different from typically developing children. When children enter kindergarten or other structured settings, they struggle with fine motor activities such as colouring, cutting with scissors and printing. Gross motor skills may be equally problematic, including activities such as hopping on one foot, learning to throw and catch a ball, riding a bicycle, using their legs to pump on a swing, hitting a ball with a bat or stick or climbing stairs with alternating feet. In later years, these differences in fine and gross motor skills limit the ability and willingness of a child with DCD to participate in physical education class and free play during school recreation, extracurricular free and organized play, and sporting activities. Their withdrawal from physical and social activity in middle and later school years can isolate them from their peers (see "Course and Developmental Challenges").

Adult participants in studies of DCD describe their inability as adolescents to participate in many life experiences such as driving a vehicle, putting on make-up or attending dance clubs (Cousins & Smyth, 2003; Kirby et al., 2011). There is persistent interference with prevocational and vocational tasks along with leisure activities into adulthood (Tal-Saban, Ornoy, & Parush, 2014). The motor impairments that are evident in childhood will persist throughout life and impact on vocational, financial and leisure pursuits, as well as interpersonal relationships (Cousins and Smyth, 2003; Fitzpatrick & Watkinson, 2003; Kirby et al., 2008; Kirby et al., 2011; Tal-Saban et al., 2014).

Screening and Diagnosis

Motor coordination difficulties can have numerous causes. These should be assessed and ruled out by conducting a complete history and physical examination, including a detailed sensory and neurological examination. All appropriate investigations should be requested, but only if needed. The presence or absence of other disorders that can cause motor incoordination needs to be considered, including genetic syndromes (e.g. Down syndrome), neurologic disorders (e.g. cerebral palsy), degenerative conditions (e.g. Duchenne's muscular dystrophy, brain tumours), musculoskeletal disorders (e.g. Legg-Perthes), physical impairments (e.g. impaired visual acuity), cognitive impairments (e.g. developmental delay), or recent injuries (e.g. traumatic brain injury) (Missiuna, Gaines, & Soucie, 2006).

The EACD Guidelines (Blank et al., 2011) suggest that a clinical developmental examination for DCD should include:

- Neuromotor status (exclusion of other movement disorders or neurological dysfunctions)
- Medical status (e.g. obesity, hypothyroidism, genetic syndromes)
- Sensory status (e.g. vision, hearing, vestibular function)
- Emotional and behavioural status (e.g. attention, self-esteem)
- Cognitive function (if there are learning difficulties at school)

Perhaps of greatest importance is for the physician to consider any indicators that would suggest that a diagnosis of DCD is not appropriate. Coordination difficulties are probably *not* attributable to DCD if there is a history of recent head injury or trauma, deterioration (child has "lost" motor skills that they previously had acquired), headaches, eye pain, or blurred vision. Physicians should also observe for the presence of increased muscle tone, fluctuating tone or significant hypotonia; asymmetry of tone or strength; musculoskeletal abnormalities; neurocutaneous lesions; Gowers' sign; ataxia or dysarthria; dysmorphic features; or the absence of deep tendon reflexes, all of which suggest a diagnosis other than DCD (Missiuna, Gaines et al., 2006).

Once medical and neurological explanations for the child's clumsiness have been ruled out, further assessment may help to determine the severity of a motor coordination disorder. If DCD is suspected, there are several evaluative tools available to assess the child's motoric capabilities. These are most typically administered by occupational therapists and physiotherapists and may be available to the physician through an assessment report. Typically, children who score between the 6th and the 16th percentile on a test of motor impairment have mild impairments and are suspected of having DCD. Those scoring below the 5th percentile have quite significant motor impairment. Of interest, a study of children with DCD who were first identified by primary care physicians showed that physicians only noticed those children in their practice who had very significant motor issues, falling below the 1st percentile on a test of motor impairment (Missiuna, Gaines, et al., 2008). During administration of a standardized test, qualitative observations provide the assessor with more detail about the child's muscle tone, strength, postural control, speed, grasp patterns, ability to use hands in a dominant/assistant fashion, effort, attention and behaviour. While the results of a standardized assessment of motor impairment are not essential for the diagnosis of DCD, this type of documentation is required in some jurisdictions to justify requests for school accommodations.

Once the presence of a motor impairment has been established, it is important to determine how a child's motor difficulties are impacting on daily performance. Through interview, questionnaires and observation, it is possible to determine the impact of poor motor coordination on *self-care activities* (dressing, hygiene, eating, toileting, bathing), *play activities* (sports, extracurricular activities, playing with friends, family outings) and *school activities* (academic progress, social relationships, following classroom routines, homework). Parents are usually able to provide this information through a checklist or short interview. Parents of children 7–16 years old can complete a validated checklist called the Developmental

Coordination Disorder Questionnaire (DCD-Q '07: Wilson et al., 2009) (http://www.dcdq.ca/). The Little DCD-Q is undergoing validation in 15 countries and should be available soon for children 3–6 years of age (Wilson et al., 2014). If physicians prefer to conduct an interview themselves, a guide called "Listening for DCD" includes questions to ask parents of school-aged children (http://canchild.ca/elearning/dcd_pt_workshop/assets/identification/listening-dcd-interview-guide.pdf). If the young person is old enough to provide self-report, the physician should also gain their perspective. With increasing maturity, questions about the impact of coordination difficulties on *pre-vocational activities* (part-time jobs, volunteering), *vocational activities* (post-secondary education, job searching, independence in managing living) and *leisure activities* (socializing, school clubs, fitness, sports) are also appropriate. A well-designed screening tool is now available for use with adults called the Adult Developmental Coordination Disorder/Dyspraxia Checklist (ADC: Kirby, Edwards, Sugden, & Rosenblum, 2010) (http://www.dyslexic.org.uk/sites/default/files/Adult_DCD_Dyspraxia_Checklist.pdf).

Associated Impairments and Comorbidities

DCD has been associated with a number of other diagnosable developmental conditions, most commonly ADHD, SLI, reading disability and LD[1] (Archibald & Alloway, 2008; Dewey, Kaplan, Crawford, & Wilson, 2002; Finlay & McPhillips, 2013; Flapper & Schoemaker, 2013; Iverson, Berg, Ellertsen, & Tonnessen, 2005; Lingam et al., 2010; Missiuna, Cairney et al., 2011; Tseng, Howe, Chuang, & Hsieh, 2007). Although exact estimates of comorbidity vary from study to study depending upon the particular sampling strategy used (clinical vs. population-based), the developmental conditions examined and the diagnostic procedures that are employed (screening vs. full diagnostic battery), researchers generally agree that comorbidity is indeed quite common (Pieters et al., 2012). Moreover, children with DCD who have more than one developmental disorder seem to have a worse prognosis (Rasmussen & Gillberg, 2000) and experience more secondary social and emotional health consequences than those with DCD alone (Flapper & Schoemaker, 2013; Missiuna et al., 2014; Piek et al., 2007). Based on these collective findings, practitioners are urged to assess all children who present with attention, language or learning problems for signs of motor impairment (Blank et al., 2011; Flapper & Schoemaker, 2013; Missiuna et al., 2014). If the criteria for more than one developmental disorder are met, then more than one developmental disorder should be diagnosed (APA, 2013).

Although children with DCD – with or without comorbidities – have a higher likelihood than typically developing children of having associated social, emotional and behavioural problems, there is debate about whether these conditions are actually comorbid or if they develop as secondary consequences of the original motor condition (see "Course and Developmental Challenges"). Longitudinal studies to fully test these two possibilities have yet to be conducted and require exploration in future research (Missiuna & Campbell, 2014).

In summary, although there is an association between DCD and other diagnosable conditions, the literature has been inconclusive in providing an explanation for these comorbidities. It appears that the greater the severity of DCD, the greater the chance of finding co-occurring developmental disorders. Although some evidence suggests that DCD shares underlying mechanisms with other developmental disorders, including an increased risk of multiple disorders, other evidence suggests more complex interactions between neurobiological differences and environmental stressors are at play (see "Etiology and Mechanisms").

[1]In the United Kingdom, learning disability is the term used to refer to intellectual delay. In North America, it usually refers to a discrepancy between the child's intellectual ability and their performance in subjects such as reading, spelling or mathematics.

Course and Developmental Challenges

As children with DCD grow older, they are at greater risk than their typically developing peers for developing a number of secondary physical health, emotional health, academic and social interaction problems (Cairney, 2015a; Zwicker, Harris, & Klassen, 2013). Table 22.2 lists the numerous secondary consequences that have been documented and may be part of the developmental progression for children with DCD, along with supporting evidence from the literature. For example, there is a growing body of evidence to suggest that children with DCD lose cardiorespiratory fitness, as well as muscle power, strength and endurance over time (Li, Wu, Cairney, & Hsieh, 2011; Rivilis et al., 2011; Wu, Lin, Li, Tsai, & Cairney, 2010). Moreover, many children with DCD show progressive and significant withdrawal from participation in physical activities over time (Green et al., 2011; Poulsen, Johnson, & Ziviani, 2011). Given that regular involvement in physical activity leads to positive physical and psychological health outcomes for children and youth more broadly, there is good reason for concern when a child progressively withdraws from physical activity (Cairney, 2015b).

In addition to its impact on physical activity and health, DCD interferes with normal developmental processes in a number of other ways. It has been well documented that children with DCD are more likely to experience emotional health problems, such as anxiety and depression, relative to their typically developing peers (Campbell et al., 2012; Lingam et al., 2012; Missiuna, Cairney et al., 2014; Pearsall-Jones et al., 2011; Piek et al., 2007; Pratt & Hill, 2011). To date, a definitive causal link between DCD and mental health has yet to be established (Missiuna & Campbell, 2014); however, one possible explanation is that early difficulties with motor coordination lead children to avoid age-appropriate physical and social activities, which in turn, may impact negatively on a child's developing self-esteem and create a sense of isolation and exclusion (Cairney, Veldhuizen, & Szatmari, 2010; Cairney, Rigoli, & Piek, 2013). Ultimately, this negative spiral from poor motor coordination to social and physical consequences may place children with DCD at risk for serious mental health issues, including depression and anxiety (Cairney, Rigoli, & Piek, 2013; Cairney, Veldhuizen et al., 2010).

Children with DCD also struggle to perform at school, demonstrating difficulties in a wide array of subjects (Dyck & Piek, 2010; Lingam et al., 2010).

Table 22.2 Secondary consequences associated with developmental coordination disorder

Secondary consequences	Research evidence
Physical problems	Poor cardiopulmonary endurance (Wu et al., 2010) Negative association with body composition, cardiorespiratory fitness, muscle strength and endurance, anaerobic capacity, power and physical activity (Rivilis et al., 2011) Sleep problems (Barnett, & Wiggs, 2012) Obesity (Cairney et al., 2011) Poor daily living activities (Carslaw, 2011) Reduced activity level (Green et al., 2011) Low participation in social-physical activities (Poulsen et al., 2011)
Social and emotional problems	Depression (Campbell, Missiuna, & Vaillancourt, 2012; Lingam et al., 2012; Missiuna et al., 2014; Piek et al., 2007) Anxiety (Missiuna et al., 2014; Pearsall-Jones et al., 2011; Pratt & Hill, 2011) Loneliness (Poulsen, Ziviani, Johnson, & Cuskelly, 2008) Social withdrawal (Chen, Tseng, Hu, & Cermak, 2009) Low self-efficacy (Engel-Yeger & Hanna Kasis, 2010) Poor peer relations and self-concept (Poulsen et al., 2011); lower global self-esteem (Eggleston, Hanger, Frampton, & Watkins, 2012) Increased risk for being bullied by peers (Campbell et al., 2012)
Academic problems	Pervasive underachievement (Dyck & Piek, 2010) Increased risk of difficulties in reading and spelling (Lingam et al., 2010)

These academic challenges may sometimes be related to co-occurring difficulties in other developmental domains such as attention, speech and language, or learning (see "Associated Impairments and Comorbidities"); however, they may also be due to a lack of recognition by educators of the motor demands of curricular tasks used to teach literacy or numeracy (Missiuna, Pollock et al., 2012). Under-recognition of children with DCD within the educational system and lack of accommodation for co-ordination difficulties are contributing factors (Missiuna, Moll, King et al., 2006; Missiuna, Polatajko, & Pollock, 2015). If children become marginalized in the classroom, they may express their frustration and unhappiness in inappropriate or even destructive ways.

Together, the many potential secondary consequences of DCD create unique challenges that can generate considerable stress for the family (Ahern, 2000; Missiuna, Moll, Law et al., 2006). Since DCD is not widely acknowledged or easily diagnosed, parents may feel frustrated, uncertain and anxious (Missiuna, Moll, Law et al., 2006). As children become increasingly aware of being "different", their anxiety may translate into physical ailments (e.g. stomach aches, headaches) or behavioural problems (e.g. crying, refusing to go to school) (Dewey et al., 2002), which can add significant pressure on the family (Pratt & Hill, 2011; Stephenson & Chesson, 2008).

In summary, DCD can have a negative effect on all aspects of a child's life. Although the direct impact of the coordination difficulties may diminish as the child learns better coping skills, the physical deterioration and emotional damage caused by this pervasive disorder often lingers for a lifetime (Cairney, 2015a; Tal-Saban, Zarka, Grotto, Ornoy, & Parush, 2012). It is only through early diagnosis of DCD and implementation of an integrated management strategy that we can hope to improve the physical, mental, social and emotional outcomes for children with DCD.

Management Many types of intervention approaches have been introduced over the years for children with DCD. Based on the assumption that we could "cure" DCD, we saw the development of process-oriented or "bottom-up" approaches including sensory integration (Ayres, 1972, 1979), kinaesthetic training (Laszlo & Bairstow, 1985; Laszlo, Bairstow, Bartrip, & Rolfe, 1988) and perceptual motor intervention (Lord & Hulme, 1987a, b). These interventions reflect traditional hierarchical theories of motor development and assume that by changing underlying deficits, skilled motor performance will improve (Mandich, Polatajko, Macnab, & Miller, 2001). However, several systematic reviews on the effectiveness of these approaches have shown that they are time-consuming, produce minimal change in functional outcomes, and that there is no clear advantage of one intervention over another (Forsyth et al., 2007; Hillier, 2007; Polatajko & Cantin, 2006; Smits-Engelsman et al., 2013).

Contemporary theories, including the work of Thelen (1995) and others, propose that changes in motor performance rely on many variables that are task- and environment-specific. Therefore, newer interventions for children with DCD reflect the belief that intervention must be contextually based: they also tend to emphasize the teaching of very specific motor skills rather than underlying skill components (Rivard, Missiuna, Pollock, & David, 2012). Described in the literature as "top-down" (Mandich et al., 2001), these interventions utilize the child's cognitive processes in the learning of new movement skills (Levac, Wishart, Missiuna, & Wright, 2009). Two examples of these approaches are Neuromotor Task Training (NTT) (Schoemaker & Smits-Engelsman, 2005) and Cognitive Orientation to daily Occupational Performance (CO-OP) (Polatajko & Mandich, 2004). In NTT, learning is directed by a physical or occupational therapist who provides verbal instructions, visual prompts or physical assistance to help the child experience and learn efficient movement while completing a motor task (Schoemaker & Smits-Engelsman, 2005). In CO-OP, the child is actively involved in choosing the tasks and goals and is guided to solve the performance problem. The target is motor skill acquisition, but the child is also learning to use

cognitive strategies during task performance. Focus and attention are directed towards the difficult aspects of the task, and it is anticipated that, through the learning process, there will be generalization and transfer to other tasks where similar difficulties are encountered (Polatajko & Mandich, 2004). Promising support for task-focused interventions such as NTT and CO-OP was reported in a recent systematic review and meta-analysis (Smits-Engelsman et al., 2013). These treatment approaches focus on the individual child who has DCD and require trained therapists to intervene. There are several reasons why it is also important to think about management of children with DCD that extends beyond the child to encompass the family, the school, and the community.

When one thinks about the overall management of a child with DCD across the lifespan, one must consider that DCD is *a learning-based motor problem* (Missiuna et al., 2015). As has been described (see "Symptom Presentation" and "Developmental Progression"), children with DCD show inconsistent motor performance because they have difficulty with motor learning, as well as the transfer and generalization of skills (Polatajko & Mandich, 2004). Given these kinds of problems, any approach to management also needs to include working with the child in the setting in which the challenges are occurring (e.g. school setting), sharing knowledge about DCD with the key adults in the child's environment such as families and educators, and providing the accommodations necessary to support the child's successful participation in school, at home and in the community (Missiuna et al., 2015). Indeed, the EACD Guidelines emphasize the importance of helping the child learn specific motor tasks in the environments in which the task is required (Blank et al., 2011).

Whereas the aforementioned interventions target individual children, the reality in many jurisdictions is that rehabilitation services are not available for children with DCD, and when they do exist, waitlists are prohibitively long (Deloitte & Touche 2010; Peters, Henderson, & Dookun, 2004). Given such resource constraints, and recognizing the relatively high prevalence of DCD, an argument has been put forth in favour of developing population-based interventions, particularly for children with DCD who are school-aged (Missiuna et al., 2015). In one population-based approach called "Partnering for Change" (Missiuna, Pollock, Campbell, et al., 2012; Missiuna, Pollock, Levac et al., 2012), intervention services are offered in progressively intensive tiers based on the child's need and his/her response to the intervention. Tier 1 services use principles of health promotion and are offered to facilitate motor skill development for all children in a given setting (such as a classroom). Tier 2 services are targeted to small groups of children who are experiencing motor coordination challenges, despite being exposed to services in Tier 1; some of these children may have DCD, but others may be having difficulties for other reasons (e.g. lack of experience). Children's response to Tier 2 interventions provides an opportunity to distinguish between these two groups of children and determine need. Finally, Tier 3 services include individualized strategies for students who probably do have DCD and who are only able to participate in school successfully with specialized supports and accommodations. In addition to being needs-based, the Partnering for Change approach specifically targets key adults in the child's environment by ensuring that therapists share knowledge about the recommendations that they provide to educators and families so that they may use successful strategies throughout the child's day (Missiuna, Pollock, Levac, et al., 2012). Thus, the Partnering for Change approach adopts a *chronic disease management perspective* that acknowledges the life-long nature of DCD.

With respect to the management of DCD, primary care physicians are often the first health professional contacted by families of children with DCD and could be the ones to facilitate earlier identification of this neurodevelopmental health condition (Gaines, Missiuna, Egan, & McLean, 2008). Many primary care physicians are unfamiliar with DCD (Wilson, Neil, Kamps, & Babcock, 2013); however, evidence-based online educational materials are available that provide guidance as to how to screen for, diagnose, manage, and monitor

children with DCD (http://machealth.ca/programs/developmental_coordination_disorder/default.aspx). Carslaw (2011) recently stressed the importance of physicians being aware of the long-term impact of DCD and of practical strategies that can be suggested to families in order to create a supportive environment for their child with DCD.

Prognosis and Outcome The prognosis for children with DCD really comes down to – "it depends". Like diabetes, DCD is a chronic health condition that needs to be actively monitored to recognize the onset of, and hopefully to prevent, secondary physical and mental health consequences. If children are identified early and receive accommodations that support independence and successful participation, then the prognosis should be positive. It is usually not the poor coordination that is the long-term problem: DCD serves as a primary stressor that – without management – can precipitate internalizing disorders such as depression and anxiety (Cairney et al., 2013). Similarly, if the motor coordination difficulties result in reduced participation in physical activity, children may become overweight or obese and physical fitness may deteriorate (Cairney, 2015b; Cairney & Veldhuizen, 2013).

Linking families to education is one of the most important ways in which the physician can provide support and improve outcomes. A website hosted by *CanChild* Centre for Childhood Disability Research (www.dcd.canchild.ca) contains an evidence-informed online module that guides families through the journey of supporting a child with DCD. What DCD looks like, understanding their child's difficulties, strategies for success and ways of advocating for their child at school and in the community are communicated clearly through user-friendly resources available in many languages. This website is also appropriate for educators of children from preschool until secondary school. Beginning with secondary school and post-secondary education, the Movement Matters website (www.movementmattersuk.org) has very helpful information outlining the characteristic presentation and difficulties of adolescents with DCD. This website contains specific suggestions of useful and feasible accommodations that will enhance the likelihood of successful completion of high school and strategies on how to prepare to enter the job market. Employers can find helpful tips to make the interview process and the job experience a positive one for adults with DCD.

Ultimately, the successful management of DCD does not rest on "curing" or "fixing" the underlying motor learning difficulty. Rather, children with DCD will grow into adults who have DCD. Based on current evidence, what appears likely to make the most difference in the ultimate trajectory of children with DCD is the early recognition and identification of their difficulties, the provision of supports to promote functioning and participation in multiple environments (i.e. home, school and community), and a focus on building the capacity of the child and key adults in his/her life to know how to successfully manage DCD over the long term. By empowering children and families to successfully advocate for themselves, it is hoped (though not yet proven) that a positive prognosis and trajectory will be achieved.

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Part X

Externalizing Disorders: Elimination Disorders

Enuresis

23

Patrina Caldwell and Alexander von Gontard

Initial Overview of the History of the Diagnosis of Enuresis in the DSMs

The Diagnostic and Statistical Manual of Mental Disorders (DSM) was developed by the American Psychiatric Association to clarify the nomenclature for psychopathology and to develop standard criteria for classifying mental disorders and for research.

When DSM-1 was published in 1952, "enuresis" was considered a psychopathology, and the psychodynamic approach assumed that enuresis was due to unresolved unconscious conflicts (American Psychiatric Association [APA], 1952). In 1968, DSM-2 classified "enuresis (special symptom not elsewhere classified)" as a psychophysiological symptom manifestation of psychopathology (APA, 1968). DSM-3 (1980) changed the term to "functional enuresis," emphasizing the exclusion of known physical etiology and focused on symptom description, defining enuresis as "the involuntary voiding of urine at least twice a month for children between the age of 5 and 6 years and once a month for older children" (APA, 1980). It used the term "enuresis" to define both incontinence during sleep and during waking hours and used the term "primary enuresis" to define children who have not yet attained urinary continence and "secondary enuresis" for those whose urinary incontinence have been preceded by at least 1 year of continence. It also acknowledged that the majority of children do not have a coexisting mental disorder, although the prevalence of coexisting mental disorders is greater in enuretics than in the general population. DSM-4 (1994) added that the urinary incontinence could be either involuntary or intentional. It changed the criteria for incontinence frequency to twice a week for at least three consecutive months (in a child of at least 5 years or equivalent developmental level) and added "the presence of clinically significant distress or impairment in social, academic (occupational), or other important areas of functioning" as an alternative criterion for the diagnosis (APA, 1994). The diagnostic criteria for DSM-5 is very similar to DSM-4 (APA, 2013).

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Presentation of the Current Diagnostic Criteria in DSM-5

Diagnostic criteria 307.6 (F98.0)

The following is the current diagnostic criteria of enuresis in DSM-5 (APA, 2013):

A. Repeated voiding of urine into bed or clothes, whether voluntary or intentional.
B. The behavior is clinically significant as manifested by either a frequency of at least twice a week for at least three consecutive months or the presence of clinically significant distress or impairment in social, academic (occupational), or other important areas of functioning.
C. Chronological age is at least 5 years (or equivalent developmental level).
D. The behavior is not attributable to the physiological effects of a substance (e.g. a diuretic, an antipsychotic medication) or another medical condition (e.g. diabetes, spina bifida, a seizure disorder).

Specify whether:

Nocturnal only:	Passage of urine only during nighttime sleep
Diurnal only:	Passage of urine during waking hours
Nocturnal and diurnal:	A combination of the two subtypes above

Comment on the DSM-5 Diagnostic Criteria for Enuresis

There are various international diagnostic criteria for enuresis: the American Psychiatric Association's Diagnostic and Statistical Manual of Mental Disorders as discussed above (APA, 2013), the World Health Organization (WHO)'s International Classification of Diseases (ICD) (World Health Organization, 2008), the International Classification of Sleep Disorders (ICSD) produced by the American Academy of Sleep Medicine (2005), and the International Children's Continence Society (ICCS)'s "Standardization of terminology of lower urinary tract function in children and adolescents" document produced by the ICCS (Austin et al., 2014).

As the ICCS's standardization of terminology document was developed by a panel of experts in the field of continence and is based on an extensive review of the literature and consensus by the ICCS's board of global multidisciplinary professionals involved with lower urinary tract function and malfunction in children, the authors believe that this is the most evidence based and relevant classification for enuresis. A comparison will therefore be made between the ICCS and DSM-5 diagnostic criteria for enuresis.

DSM-5 uses the umbrella term "enuresis" to include the passage of urine during nighttime sleep and during waking hours and notes that the "nocturnal-only subtype of enuresis "is "sometimes referred to" as "monosymptomatic enuresis," the "diurnal-only subtype of enuresis" "may be referred to" as "urinary incontinence," and the "nocturnal and diurnal subtype" "is also known" as "non-monosymptomatic enuresis" (APA, 2013).

In contrast, ICCS use the term "enuresis" exclusively for intermittent passage of urine during sleep and use a separate term "daytime urinary incontinence" for urinary incontinence when awake. ICCS would classify those who have both daytime wetting and nighttime wetting with a dual diagnosis of "daytime urinary incontinence and nocturnal enuresis." ICCS subgroups the nighttime wetting into monosymptomatic (night wetting without lower urinary tract symptoms) and non-monosymptomatic (night wetting with concomitant lower urinary tract symptoms, irrespective of the presence or absence of daytime urinary incontinence) as the pathogenesis and management of these two subtypes of nighttime wetting differ. This subtle difference means that the condition of nighttime wetting with lower urinary tract symptoms (such as urgency and frequency) but without concomitant daytime wetting would be classified as "monosymptomatic enuresis" by DSM-5 and "non-monosymptomatic enuresis" by ICCS (Austin et al., 2014).

The definitions for secondary enuresis also differ between DSM-5 and ICCS. DSM-5 defines secondary enuresis as wetting which "develops after a period of established urinary continence" without specifying a time period, whereas ICCS specifies a previous dry period of at least 6 months for defining secondary enuresis.

DSM-5 defines enuresis as wetting at least twice a week for at least three consecutive months or the presence of clinically significant distress or impairment in social, academic (occupational), or other important areas of functioning. In contrast, the ICCS defines the "disorder" of enuresis and daytime urinary incontinence by a minimum frequency of once per month (which is less frequent than the DSM-5 definition). If it occurs less than once a month, it is considered to be a symptom, but not a disorder. Similar to DSM-5, the ICCS definition requires a duration of at least three consecutive months. Also, ICCS quantifies the frequency of enuresis as frequent (>4 per week) or infrequent (<4 per week) (Austin et al., 2014). There is no mention of the impact on quality of life in the ICCS definition, as this can be subjective and difficult to quantify.

Other subtle differences between DSM-5 and ICCS include the minimum age required for a diagnosis. According to the ICCS definition, the minimum chronological age of 5.0 years is decisive, while DSM-5 adds the "equivalent developmental age" as an alternative. As many children over 5.0 years of age with special needs such as intellectual disability or autism spectrum disorder may have a much lower developmental age, these patients would not fulfill the criteria of a DSM-5 diagnosis and may be excluded from medical care (von Gontard, 2013a, 2013b).

Regarding daytime incontinence, DSM-5 only describes two groups, urge incontinence and voiding postponement, whereas ICCS identifies many conditions that cause daytime urinary incontinence.

Because of the discrepancies between ICCS and DSM-5 definitions, we will use the term "enuresis" to define nighttime wetting and "daytime urinary incontinence" to define wetting when awake throughout this chapter.

Research Review

Etiology

The understanding about the cause of urinary incontinence has changed over time from a psychopathologic and psychodynamic focus to a multifactorial focus with an emphasis on physiology, psychology, and genetics. Urinary incontinence can be continuous or intermittent. As continuous incontinence is usually associated with anatomic or physiological abnormalities which is excluded in the DSM-5 definition, the focus of this chapter will be on intermittent urinary incontinence. As enuresis and daytime urinary incontinence are both included in the DSM-5 definition of "enuresis" but are separate conditions, they will be discussed separately.

Enuresis

The etiology of nighttime wetting is not fully understood, but is thought to be a complex interaction of multiple physiological, psychological, and environmental factors. Proposed causes include delayed maturation of the micturition process, central nervous system immaturity and developmental delay (Järvelin, 1989), altered bladder physiology, psychiatric and behavioral disturbances, environmental factors, and genetic factors (von Gontard, Schaumburg, Hollmann, Eiberg, & Rittig, 2001). It is proposed that the symptom of nighttime wetting relates to the interplay of three elements: defective sleep arousal, lack of inhibition of bladder emptying during sleep, and nocturnal urine production (Butler & Holland, 2000; Koff, 1996). Studies have demonstrated that the arousal response to the sensation of a full bladder is defective during sleep in many enuretic children (Wolfish, Pivik, & Busby, 1997). Some also have a reduced bladder capacity and wet when their urine volume exceeds their bladder capacity during sleep. Children who have high overnight urine production (nocturnal polyuria) caused by excessive evening fluid intake or excessive urine production secondary to alterations in the circadian secretion of antidiuretic hormone by the pituitary

gland may also wet if they are unable to wake when their bladder is full. A subgroup of enuretic children secrete a lower level of antidiuretic hormone during sleep (compared with those without night wetting), resulting in an excessive production of relatively dilute urine (Nørgaard, 1991; Rittig, Knudsen, Norgaard, Pedersen, & Djurhuus, 1989). Other conditions which impact on enuresis include sleep-disordered breathing (Brooks & Topol, 2003), constipation (Loening-Baucke, 1997; McGrath, Caldwell, & Jones, 2008; O'Regan, Yazbeck, Hamberger, & Schick, 1986), attention deficit/hyperactivity disorder (ADHD) (von Gontard & Equit, 2014), and other comorbid disorders (von Gontard, Baeyens, Van Hoecke, Warzak, & Bachmann, 2011).

Daytime Urinary Incontinence

Studies have shown that genetic factors play a role in the etiology of daytime urinary incontinence. In a population-based study of over 10,000 families, the odds ratios for daytime urinary incontinence two or more times per week in the child were 3.28 times higher if the mother and 10.1 times higher if the father had a history of daytime urinary incontinence (von Gontard, Heron, & Joinson, 2011).

Delayed development, difficult temperament, and maternal depression/anxiety were associated with later attainment of daytime continence in a cohort of 10,000 children aged 4–9 years (Joinson et al., 2008). These findings suggest that pre-existing risk factors and stressors can affect attainment of continence, although continence was not influenced by early or intensive toilet training in general (Largo, Molinari, Siebenthal, & Wolfensberger, 1996).

Daytime urinary incontinence is caused by a heterogeneous group of disorders. In some, genetic etiological influences predominate; in others environmental risk factors are decisive. Therefore, each of the subgroups will be considered separately.

Urge incontinence is caused by spontaneous detrusor contractions during the filling phase of the bladder, which are not sufficiently inhibited by the central nervous system (Franco, 2007a, 2007b). Linkage studies demonstrate a possible dominant mode of transmission and linkage to chromosome 17 (Eiberg, Shaumburg, von Gontard, & Rittig, 2001). Behavioral symptoms and disorders are less common in urge incontinence than in other subtypes of daytime urinary incontinence such as *voiding postponement*, which can be due to an acquired habit or may be a symptom of oppositional defiant disorder (ODD) (Kuhn, Natale, Siemer, Stoeckle, & von Gontard, 2009).

Etiological data on other types of daytime urinary incontinence are not robust and are often based on anecdotal reports (*Management of disorders of bladder and bowel control in childhood*, 2006). *Dysfunctional voiding* rarely occurs in other family members. It is usually an acquired habit but can be associated with severe psychological disturbances, familial stressors, and child neglect. *Underactive bladder* can evolve from overactive bladder, voiding postponement, and dysfunctional voiding. *Stress incontinence* (involuntary leakage or urine with effort or physical exertion) is extremely rare in children, in contrast to adults, and may be related to poor pelvic floor control. *Vaginal reflux* is caused by urine refluxing into the vagina during voiding and leaking out afterward. *Giggle incontinence* is caused by a sudden transient loss of muscle tone, is related to cataplexy and narcolepsy, and can be familial (Austin et al., 2014).

Symptom Presentation

Enuresis

According to the ICCS, enuresis (or nocturnal enuresis) describes any intermittent incontinence occurring during sleep (including daytime naps). Enuresis can be further subtyped according to onset (as primary or secondary, if the child had been previously dry) and the presence or absence of lower urinary tract symptoms (monosymptomatic or non-monosymptomatic enuresis).

Primary Versus Secondary Enuresis

Primary enuresis means the child has not attained at least 6 months of nighttime continence. Primary enuresis is more common than secondary

enuresis in younger children. In a large population-based study, 11.0% of 5-year-old children had primary and 4.7% secondary enuresis; at the age of 8 years, the rates were 3.3% and 4.1%, respectively (Fergusson, Horwood, & Shannon, 1986). Primary enuresis was influenced by a family history of enuresis, as well as early developmental factors, but not by psychosocial factors (Fergusson et al., 1986).

Secondary enuresis is a relapse in night wetting after a dry period of at least 6 months, irrespective of whether continence was achieved spontaneously or as a result of treatment. This dry interval can occur at any age. Children who attain nighttime continence after the age of 5 years have a greater risk for a later relapse (Fergusson, Horwood, & Shannon, 1990). Population-based studies have shown that children with secondary enuresis have experienced more stressful life events, which can precipitate the relapse (Fergusson et al., 1990; Järvelin, Moilanen, Vikeväinen-Tervonen, & Huttunen, 1990). The most important single life event was separation/divorce of parents, and the most vulnerable age for secondary enuresis was 5 and 6 years, with the highest level of weighted life events (Järvelin et al., 1990).

The somatic symptoms of primary and secondary enuresis are quite similar (Robson, Leung, & Van Howe, 2005). In Robson's series, children with secondary enuresis had higher rates of constipation but did not differ regarding lower urinary tract symptoms, uroflowmetry patterns, post void residual bladder volumes, urinary tract infections, and a variety of other parameters (Robson et al., 2005).

Monosymptomatic Versus Non-monosymptomatic Enuresis

The diagnosis of monosymptomatic and non-monosymptomatic enuresis are based entirely on clinical assessment (history and voiding diaries) (*Management of disorders of bladder and bowel control in childhood*, 2006).

Children who wet during sleep and do not have any lower urinary tract symptoms have *monosymptomatic* enuresis. Monosymptomatic enuresis is more common than non-monosymptomatic and affects 68.5% of children with enuresis (Butler & Heron, 2006). Presenting symptoms are usually poor sleep arousal and increased urine volumes at night (polyuria). The typical child with monosymptomatic enuresis is extremely difficult to wake and passes a large amount of urine onto the bed without waking up. In contrast, bladder function during the day is completely normal.

Children with enuresis and any lower urinary tract symptoms have *non-monosymptomatic* enuresis. Although they can also have arousal difficulties and polyuria, they experience similar lower urinary tract symptoms to children with daytime urinary incontinence (Franco, von Gontard, & De Gennaro, 2013). Symptoms and signs include urgency, frequency, voiding postponement, or dysfunctional voiding. Also, urinary tract infections, constipation, and fecal incontinence can coexist. In a large epidemiological study, children with non-monosymptomatic enuresis had more behavioral symptoms than those with the monosymptomatic type (Butler & Heron, 2006). Other differences included increased daytime urinary and fecal incontinence, multiple episodes of nighttime wetting, and the need for prompting to go to the toilet during the day for those with non-monosymptomatic enuresis (Butler & Heron, 2006).

Daytime Urinary Incontinence

The three most common ICCS-defined subtypes of intermittent daytime urinary incontinence are urge incontinence (overactive bladder), daytime urinary incontinence with voiding postponement, and dysfunctional voiding. Rare forms of daytime urinary incontinence include underactive bladder, stress incontinence, vaginal reflux, giggle incontinence, and extraordinary daytime urinary frequency (Austin et al., 2014).

Children with *urgency (or urge) incontinence* are characterized by urinary urgency, which can arise suddenly and intensively, frequent micturition (more than seven voids per day with short intervals between voids), and smaller than expected voided volumes. Incontinence episodes are more common during the afternoons,

increasing with tiredness. To stop wetting, children use holding maneuvers such as contracting the pelvic floor muscles, pressing the thighs together, holding the abdomen, jumping from one leg to the other, sitting on their heel, or squatting with the heel pressed against the perineum (curtsey sign). Psychological symptoms are less common than in other subtypes such as voiding postponement (Kuhn et al., 2009). Urge incontinence can be associated with vulvovaginitis, peri-genital dermatitis, and urinary tract infections.

Typical symptoms of *daytime urinary incontinence with voiding postponement* are a habitual deferral of micturition in certain situations (school, play, reading, television). With increasing filling of the bladder with postponement, a feeling of urgency can arise, and holding maneuvers are instituted (the same as in urge incontinence) until wetting cannot be avoided. Some children learn to restrict fluid intake in an attempt to avoid incontinence. Children with voiding postponement often void less than five times per day and may also have constipation and fecal incontinence. Children with voiding postponement have the highest rate of behavioral disorders (Kuhn et al., 2009; Zink, Freitag, & von Gontard, 2008), with the most common being oppositional defiant disorder (ODD) .

Dysfunctional voiding (discoordination of the bladder and the sphincter, with intermittent sphincter overactivity during voiding) is characterized by repeated abdominal straining at the beginning and during micturition (Chase, Austin, Hoebeke, & McKenna, 2010). The urine flow is interrupted leading to staccato or interrupted curves on uroflowmetry. Dysfunctional voiding is associated with incomplete bladder emptying, urinary tract infections, vesicoureteral reflux, stool retention, constipation, and fecal incontinence.

Clinical symptoms of *stress incontinence* are leakage of small amounts of urine with increased intra-abdominal pressure (coughing, sneezing, during play and sports). In *giggle incontinence*, the wetting is initiated by laughing (not by increased intra-abdominal pressure) and often cannot be stopped until the bladder is empty. Children with *underactive bladder* need raised intra-abdominal pressure to initiate, maintain, or complete voiding. They usually have a large bladder, void infrequently (less than five times per day), and do not completely empty their bladder when they void, with large residual urine volumes. They exhibit an interrupted uroflow pattern. Girls with *vaginal reflux* leak moderate amounts of urine in their pants shortly after voiding. They often void with their legs together or crossed. In *extraordinary daytime urinary frequency,* there is usually a sudden onset of urinary frequency with small voided volumes limited to when the child is awake. Urgency can also be a symptom. The course is usually benign and self-limiting and can last from a few weeks up to 6 months.

Children with Both Enuresis and Daytime Urinary Incontinence

According to ICCS, if a child has both daytime urinary incontinence and enuresis, two diagnoses should be given. These children have significantly more conduct and oppositional problems than in those with enuresis alone (Joinson, Heron, Emond, & Butler, 2007).

Associated Impairments and Comorbidities

Children with enuresis or daytime urinary incontinence are often distressed and have a reduced quality of life (Bachmann et al., 2009; Deshpande, Craig, Smith, & Caldwell, 2011). In addition, comorbid behavioral disorders are more common in children with incontinence than in their continent peers, as many population-based studies have shown (von Gontard, Baeyens et al., 2011).

ADHD is the most common comorbid disorder in children with *enuresis*. In a population-based study of 1379 6-year-old children, 9.4% with enuresis had clinically relevant ADHD symptoms, compared to 3.4% of non-wetting children (von Gontard, Moritz, Thome-Granz, & Freitag, 2011). Enuresis also tends to persist for longer in children with ADHD (Baeyens et al., 2004, 2005) because of poorer compliance and treatment outcomes (Crimmins, Rathbun, & Husmann, 2003).

Of the subgroups of enuresis, children with *secondary enuresis* have the highest rates of comorbid psychological disorders, with up to 40% in population-based and 75% in clinical studies (Feehan, McGee, Stanton, & Silva, 1990; von Gontard, Baeyens et al., 2011; von Gontard, Mauer-Mucke, Plück, Berner, & Lehmkuhl, 1999).

In a British population-based study of over 8000 children aged 7½ years, those who had enuresis only were affected by separation anxiety (8.0%), social anxiety (7.0%), specific phobia (14.1%), generalized anxiety (10.5%), depression (14.2%), ODD (8.8%), conduct disorders (8.5%), and ADHD (17.6%) (Joinson et al., 2007). Those with non-monosymptomatic enuresis (Butler & Heron, 2006) and combined day and night wetting have even more comorbid disorders than those with monosymptomatic enuresis (Joinson et al., 2007) with ADHD affecting 24.8%, ODD 10.9%, and conduct disorders 11.8% of children with combined day and night wetting.

Overall, children with daytime urinary incontinence have higher rates of associated medical disorders such as urinary tract infections and vesicoureteral reflux, as well as psychological disorders such as ADHD (24.8%) and other externalizing disorders such as conduct disorders (11.8%) and ODD (10.9%) than children with enuresis (von Gontard, Baeyens et al., 2011). 20–30% of children with enuresis, 20–40% with daytime urinary incontinence, and 30–50% with encopresis have clinically relevant comorbid behavioral disorders (von Gontard, Baeyens et al., 2011). A recent comprehensive review demonstrated that children with daytime urinary incontinence are much more likely to be affected by ADHD than children with enuresis (von Gontard & Equit, 2014). Again, compliance and treatment outcomes are worse if both daytime urinary incontinence and ADHD are present (Crimmins et al., 2003).

Course and Prognosis

Enuresis

In one epidemiological study, the spontaneous remission rate for enuresis with no treatment was reported as 13–15% per year (Forsythe & Redmond, 1974). In another study where children were offered treatment, the incidence of enuresis reduced from 7% to 0.6% for girls and 12% to 0.3% for boys at aged 7 and 17 years, respectively (Hellstrom, Hanson, Hansson, Hjalmas, & Jodal, 1995). The risk for enuresis in adulthood is significantly higher (up to eightfold) in those who had experienced childhood enuresis. The overall prevalence of enuresis (at least once per month) in those aged 33–60 years was 0.4% for women and 0.3% for men and for enuresis of less than once per month was 1.7% for both genders. In those who had experienced enuresis in childhood, these rates increased to 3.0% for women and 2.5% for men who wet at least once per month and 7.8% for both genders who wet less than once per month (Hublin, Kaprio, Partinen, & Koskenvuo, 1998).

The severity of enuresis has been shown to influence the long-term outcome. Children with infrequent enuresis (frequency of less than three wet night per week) were more likely to become dry than those who wet every night (Yeung, Sreedhar, Sihoe, Sit, & Lau, 2006). A British longitudinal population-based study analyzed the effects in women who experienced enuresis at aged 6 years. At the age of 48 years, women who had previously experienced enuresis had a higher rate of stress incontinence (66.7% vs. 49.8%), urge symptoms (33.3% vs. 22.3%), and severe incontinence (22.2% vs. 76%) than those who had no incontinence as children (Kuh, Cardozo, & Hardy, 1999). In one study from Hong Kong, 2.3% of adolescents and adults remained enuretic at age 16–40 years (Yeung et al., 2004).

Daytime Urinary Incontinence

Daytime urinary incontinence is not only a heterogeneous condition but can take different courses or trajectories. Based on a longitudinal cohort of nearly 11,000 children aged 4 ½ to 9 ½ years, four different trajectories could be identified: 86.2% of children took a normative course with negligible daytime wetting, 6.9% took longer to become dry (delayed), 3.7% had a relapse after attaining continence, and 3.2% were persistently wet (Heron, Joinson, Croudace, & von Gontard, 2008). The data regarding daytime urinary incontinence from Hellström's study where children were offered

treatment showed the incidence of daytime urinary incontinence reduced from 6% to 3.6% for girls and 4% to 0.3% for boys at aged 7 and 17 years, respectively.

Developmental Challenges

Children usually achieve daytime continence before nighttime continence. As attainment of bladder control is a developmental milestone, developmental factors (such as intellectual and physical capacity) can impact on bladder training and attainment of continence. For example, about 94% of the general childhood population achieve daytime urinary continence by 4 years of age compared to only 54.3% of children with cerebral palsy (Brazelton, 1962; Roijen, Postema, Limbeek, & Kuppevelt, 2001) and about 85% of the general population achieve nighttime continence by 4 years compared to only 34.4% of those with cerebral palsy. Delayed initiation of toilet training beyond the age of 2 years (after adjusting for developmental delay) was significantly associated with persistent or delayed wetting, as well as with relapse (Joinson et al., 2009).

Treatment

The specific identification of the subtypes of incontinence has a major impact on the sequence and types of treatment, especially when several different subtypes co-occur. All children should be offered urotherapy (a term which describes conservative interventions such as educating the child and family about the condition, general advice about correct voiding posture, regular voiding, optimizing fluid intake, ensuring adequate sleep and treating constipation), as this helps improve bladder function. Any constipation and fecal incontinence should be treated first, as this alone can lead to resolution of urinary incontinence (Loening-Baucke, 1997). If both daytime urinary incontinence and nocturnal enuresis exist, daytime symptoms should be treated before night symptoms.

Enuresis

ICCS recommends that treatment for enuresis be offered to children over the age of 5 years, according to the definition for enuresis (Neveus et al., 2010). A thorough history, physical examination, and limited investigation (including a bladder diary of fluid intake and urine output) are essential for differentiating the subtypes of enuresis. As there is an increased risk for adverse life events and coexisting psychological disorders in children with secondary enuresis, special attention should be paid to this group in the assessment and management, and all comorbid disorders should be addressed. Otherwise, the treatment of primary and secondary enuresis is exactly the same. In monosymptomatic enuresis, treatment can focus primarily on the wetting episodes during sleep, but in non-monosymptomatic enuresis, the lower urinary tract symptoms should be treated before the night wetting. If enuresis persists, then it is treated according to evidence-based recommendations (Nunes, O'Flynn, Evans, & Sawyer, 2010; Neveus et al., 2010). Specific treatments for enuresis include alarm training, desmopressin, tricyclic antidepressants, and other therapies including combination therapy.

Alarm Training is the most effective long-term treatment and is a first-line therapy for enuresis (Glazener, Evans, & Peto, 2005). Enuresis alarms are wetness sensors that are placed on the bed (bell and pad alarm) or worn in the child's underpants (body worn alarm) that signal and wakes the child when they wet. Alarms work by operant conditioning the child to wake in response to the sensation of a full bladder or to "hold on" and withhold voiding if they do not need to void during sleep (Glazener et al., 2005). Alarm training usually takes several weeks to start working and should be continued until the child achieves 14 consecutive dry nights (usually after 2–3 months) (Neveus et al., 2010). About two thirds of children become dry during alarm training, and nearly half remain dry after stopping treatment. It can be used more than once (Gim, Lillystone, & Caldwell, 2009), and relapse can be reduced by "overlearning" where the child has a large drink at night

while continuing to wear the alarm after they initially achieve success, to reinforce learning to wake to void during sleep (Glazener et al., 2005; Young & Morgan, 1972). Although alarm training is relatively inexpensive and potentially curative, it requires supportive parents and a motivated child, and treatment failure commonly occurs.

Desmopressin is a synthetic analogue of antidiuretic hormone (arginine vasopressin) which is normally secreted by the pituitary gland during sleep. It exerts an antidiuretic effect by increasing the reabsorption of water in the collecting tubules of the kidneys and reduces the volume of urine produced at night. Desmopressin is also considered a first-line therapy for enuresis and has a more rapid effect than alarm training, although this response is not sustained. Desmopressin reduces the frequency of wetting, but only 20% will become completely dry on treatment (Glazener & Evans, 2002), with 60–80% relapsing on discontinuation the medication. Because of its rapid effect, desmopressin is particularly useful for short-term needs such as for sleepovers and overnight school excursions, or when alarm training is not possible. Desmopressin can also be used if alarm therapy is ineffective and can be combined with other therapies if monotherapy is ineffective (Glazener & Evans, 2002). Desmopressin is available as a tablet, lysate (melt), or nasal spray (although the nasal spray is not available in some countries due to their increased risk for side effects) and is taken before bed. As desmopressin can increase the risk of hyponatremia and water intoxication, minimizing fluid intake at night after taking the medication is essential.

Tricyclic Antidepressants (such as imipramine and amitriptyline) have been used to treat enuresis since the 1970s. Although the mechanism of action is unknown, tricyclics are thought to exert an anticholinergic effect on the bladder and may possibly also improve arousability by altering central adrenergic imbalances (Glazener, Evans, & Peto, 2003). Tricyclics reduce the frequency of wetting, and about 20% become dry on treatment, with most relapsing when the medication is discontinued (Glazener et al., 2003). Because of concerns regarding the adverse effects of tricyclics (such as cardiotoxicity and hepatotoxicity with overdose), tricyclic use requires close supervision. Given the availability of safer treatment options (such as alarm training and desmopressin), tricyclics are used as third-line therapy in enuresis.

Anticholinergics and Other Therapy Anticholinergic medications (such as oxybutynin and propiverine) are commonly used to treat overactive bladder. Although it is ineffective as monotherapy for treating enuresis, it is sometimes used in combination with other therapies such as alarm training or desmopressin for non-monosymptomatic enuresis or resistant enuresis (Deshpande, Caldwell, & Sureshkumar, 2012). Anticholinergics inhibit bladder contractions and can improve the compliance of the bladder. Adverse effects such as behavioral changes, facial flushing, reduced sweating, dry mouth, constipation, and urinary retention are not uncommon for the older anticholinergics such as oxybutynin. Although newer, bladder-specific anticholinergics with fewer side effects are available (such as tolterodine and solifenacin), current evidence supporting pediatric use is limited.

Daytime Urinary Incontinence

The same basic principles as in the treatment of enuresis should be followed for *daytime urinary incontinence.* A thorough assessment should be conducted to determine the subtype of daytime urinary incontinence, and all comorbid disorders should be addressed.

The main focus in the treatment of *urge incontinence* is a symptom-oriented cognitive-behavioral approach aimed at a conscious control of the urge without the use of holding maneuvers (Vijverberg, Elzinga-Plomp, Messer, Van Gool, & De Jong, 1996). Children are instructed to record when they feel an urge and go to the toilet immediately without using holding maneuvers. They also record if their pants were dry or wet. It is important that children are allowed to access the toilet whenever they feel an urge, especially at school. This simple approach is sufficient for many children.

Others may require the combination of training and anticholinergics (see previous about anticholinergic use).

A symptom-oriented approach is also indicated for *voiding postponement*. The main component is regular timed voiding seven times a day in a relaxed manner. Each voiding attempt and wetting episode is recorded. Often children need reminders to go to the toilet. To increase motivation, a simple token system with positive reinforcement or reminders on an alarm watch or mobile phone can be helpful. As many children are affected by ODD, additional child psychiatric assessment and treatment is often needed.

Treatment of *dysfunctional voiding* includes increasing motivation, cognitive-behavioral elements, sphincter relaxation, and urotherapy (Chase et al., 2010). Medication is usually not indicated. The most specific and effective treatment for dysfunctional voiding is biofeedback, either with uroflowmetry or pelvic floor EMG (Chase et al., 2010).

Vaginal reflux is treated by altering voiding posture (usually by straddling the toilet backward). *Giggle incontinence* is treated with a classical conditioning paradigm (Elzinga-Plomp et al., 1995) or with the stimulant methylphenidate (due to the overlap with cataplexy and narcolepsy) (Sher & Reinberg, 1996). *Stress incontinence* is treated by pelvic floor physiotherapy or imipramine. Children with *underactive bladder* require intensive training and often intermittent catheterization. All of these rare disorders do require specialized treatment (*Management of disorders of bladder and bowel control in childhood*, 2006).

In therapy-resistant *daytime urinary incontinence*, structured group therapies have been successful (Equit, Sambach, Niemczyk, & von Gontard, 2014). These combine counseling, provision of information, relaxation techniques, cognitive-behavioral, play and group therapy approaches.

Outcome

Although effective treatment is available, the majority of families do not seek treatment for their child's enuresis or daytime urinary incontinence (Bower, Moore, Shepherd, & Adams, 1996). Persistent enuresis and daytime urinary incontinence is associated with a variety of negative consequences such as recurrent urinary tract infections (Hooton, 2001), higher rates of depression (Macaulay, Stern, & Stanton, 1991; Nygaard, Turvey, Burns, Crischilles, & Wallace, 2003) and sexual dysfunction (Salonia et al., 2004; Temml, Haidinger, Schmidbauer, Schatzl, & Madersbacher, 2000), and lower quality of life (Hunskaar & Vinsnes, 1991) and educational level. Self-esteem can improve with treatment (Hagglof, Andren, Bergstrom, Marklund, & Wendelius, 1998), sometimes irrespective of treatment success (Longstaffe, Moffatt, & Whalen, 2000). It is therefore imperative that children experiencing enuresis and daytime urinary incontinence be offered appropriate and effective treatment.

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Encopresis

24

Melissa DeVries

Introduction

Bowel and bladder continence is a widely accepted requirement for socialization across cultures. Although the expected age of completion of toilet training may vary somewhat across cultures, it is suggested that 80–90% of individuals achieve this milestone according to their specific cultural normative timeline (Whiting & Child, 1953). For those individuals who fail to meet or sustain this developmental accomplishment, absent organic cause, an elimination disorder diagnosis, may be considered. This chapter will discuss the history of encopresis as a biobehavioral disorder within the DSM framework, current diagnostic criteria, etiology, symptom presentation, associated impairments and common comorbidities, the typical course of the disorder and developmental challenges, diagnostic procedures and treatment, and the prognosis and outcomes for those diagnosed.

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History of Encopresis within the DSM Framework

The term encopresis is derived from the Greek word for stool, *kopros* (Philiohi, 2008). Although it has appeared in the literature since the early twentieth century (Weissenberg, 1926) as the fecal counterpart to enuresis (urinary incontinence), there existed much debate over how the term should be applied (Levine, 1975). For example, Levine noted it has been used to refer to everything from cases of severe constipation and overflow-related incontinence to those children who exhibit undergarment staining solely due to poor toileting hygiene.

Encopresis was not included in the original version of the Diagnostic and Statistical Manual of Mental Disorders (DSM; American Psychiatric Association [APA], 1952). Rather, it first appeared in the DSM-II (APA, 1968) within a section titled "special symptoms." At that time, the short list of disorders categorized within this section were described as those conditions that were manifested by a single symptom, were not the result of an organic illness or defect, and were not elsewhere classified. Among this list were "speech disturbance, specific learning disturbance, tic, other psychomotor disorder, disorder of sleep, feeding disturbance, enuresis, encopresis, cephalalgia, and other special symptom"

S. Goldstein, M. DeVries (eds.), *Handbook of DSM-5 Disorders in Children and Adolescents*,
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(pg. 48). Although encopresis was listed as a diagnosis, no descriptive information was provided about the term. It is also of note that the "special symptoms" section was separate from the section discussing behavior disorders of childhood and adolescence.

In 1980, with the publication of the third edition of the DSM, encopresis underwent major changes or elaborations as a diagnostic category. Twelve years after first appearing as a diagnostic classification, encopresis was objectively defined (MacLean & Brayden, 1992). It was defined as the repeated voluntary or involuntary defecation in inappropriate places and was categorized as a disorder with physical manifestations (Doleys, 1988; MacLean & Brayden, 1992). Additionally, encopresis was moved to the section on mental disorders usually first evident in infancy, childhood, or adolescence. The DSM-III (APA, 1980) also noted that the feces passed should be of normal or near normal consistency and that the location of passage should be deemed inappropriate within the individual's own sociocultural setting. Exclusionary criteria included cases in which there was a known organic causes for the disorder. With this edition of the DSM, encopresis was also further distinguished as either *primary encopresis* if the child had never displayed full bowel continence (for at least 12 months) before the onset of the disorder or *secondary encopresis* for when the condition occurred in children who were previously bowel continent (for at least 12 months) although even to the present day, this distinction has not been shown to have any therapeutic relevance (von Gontard, 2014).

Further diagnostic clarification was offered in the revised edition of the DSM-III (DSM-III-R; APA, 1987). It had been previously noted that the diagnosis required a minimum chronological age of 4 years; the generally accepted age expectation for bowel training to have been completed, at least within the United States (Doleys, 1988). The DSM-III-R went a step further and clarified that a diagnosis of encopresis was only appropriate in individuals with a mental age of 4 years or older. This was an important modification to the diagnostic criteria considering the developmental requirements for successful independent toileting include:

1. Adequate language skills to communicate with caregivers the need to eliminate
2. The ability from a social/emotional viewpoint to recognize the societal expectations for toileting
3. The fine motor skills necessary to manipulate clothing and toilet paper
4. The gross motor skills necessary for body posture in using the toilet appropriately
5. The cognitive skills needed to recognize the bodily indications of the need to defecate and the planning and self-control to do so appropriately (Fischel & Liebert, 2000)
6. The neurological maturation needed for sphincter control (MacLean & Brayden, 1992)

This modification likely reduced the risk of overdiagnosis in children with developmental delays or below average intellectual ability.

Although there was still no research to support the need for distinctions between *primary* and *secondary encopresis*, these divisions were carried in the DSM-III-R (MacLean & Brayden, 1992). However, other diagnostic changes were made within the DSM-III-R. The notations that sociocultural acceptability of toileting location and the consistency of stool should be taken into account were removed, as they were not diagnostically discriminatory (APA, 1987).

In 1994 when the APA published the fourth edition of the DSM, the diagnostic criteria began to more closely resemble the criteria presently used. Encopresis was defined as the repeated passage of fecal matter, regardless of intentionality, in inappropriate places, such as one's clothing or the floor). These criteria required a frequency and duration of one event per month over the course of at least 3 months in a child of at least 4 years of age (developmentally). It was also noted that diagnosis should not be assigned in the case where symptoms were directly due to organic cause, including side effect of a substance (such as laxatives or other medications) or medical condition, "except through a mechanism involving constipation" (p. 106). Two subtypes of encopresis were described: (1) with constipation

and overflow incontinence and (2) without constipation and overflow incontinence. Constipation could be demonstrated through physical exam or by history. These criteria have been continued, virtually unchanged with the more recent updates of the DSM (DSM-IV-TR; APA, 2000)(DSM-5; APA, 2013).

Current Diagnostic Criteria in the DSM-5

Encopresis as currently defined in the fifth edition of the DSM continues to be listed as an elimination disorder usually first diagnosed in infancy, childhood, and adolescence. The defining feature of the diagnosis is the repeated passage of fecal matter in inappropriate places either involuntarily or intentionally. In order to diagnose the condition, it should have occurred at least once per month for a period of at least 3 months in an individual who is 4 years of age or older (or developmentally equivalent to a child of 4 years or older). The etiology of fecal incontinence cannot be directly due to medical condition or substance use (including laxatives) except where constipation is involved. The DSM-5 notes that there are many possible causes of constipation including behavioral reasons and medical/physiological reasons. Subtypes of encopresis continue to be used in diagnosing the condition including encopresis with constipation and overflow incontinence (this requires the passage of stool less than 3 times per week) and encopresis without constipation and overflow incontinence (where there is no evidence on physical exam or by history of constipation and soiling is no more than intermittent) (DSM-5; APA, 2013).

Etiology

In the majority of cases, encopresis develops from process of repeated withholding of stool due to history of passing painful/large diameter stool. This guarding reflex, or withholding of stool, results in a state of chronic constipation (Mellon, 2012) through a pattern of escape/avoidance learning (Fischel & Liebert, 2000) just as healthy toileting behaviors are acquired by natural consequences and reinforcements delivered from the environment (Mellon, 2012).

Others have pointed to environmental causes that may lead to encopresis. For example, Mota and Barros (2008) noted that premature initiation of toilet training and stressful events occurring during toilet training could not only extend the training process but could also lead to development of encopresis. Not all studies have supported the finding that early initiation of toilet training alone increases risk of developing encopresis (Fishman, Rappaport, Cousineau, & Nurko, 2002). However, there is evidence that stressful events including harsh parental reactions to toileting experience and episodes of incontinence (Mast & Smith, 2012) can lead to encopresis. Fishman, Rappaport, Cousineau, and Nurko (2002) found that in over 400 children with encopresis, those with primary encopresis were more likely to have experienced interrupted toilet training, punishment associated with toilet training, constipation and abdominal pain during toilet training, and fear of the toilet.

It is a common misconception that encopresis results from sexual abuse victimization. Yet the prevalence of this symptom in the population of sexual abuse victims appears equivalent to the rate of encopresis within the general population and thus is not a good indicator of sexual abuse history (Brayden & Altemeier, 1989). For example, Anderson et al. (2014) retrospectively reviewed medical charts of 1280 children presenting in clinics for non-acute examination following sexual abuse allegations. They found that only 2.3% of these youth presented with symptoms of encopresis, which is consistent with the rates of encopresis noted in the general population. Encopresis, and soiling behavior in particular has, at the very least, been demonstrated as no more common in sexual abuse victims than in children with other psychiatric conditions and thus should be considered one of many dysregulated behaviors that appears in youth with psychiatric conditions and not strongly indicative of sexual abuse history (Mellon, Whiteside, & Friedrich, 2006).

Symptom Presentation

Encopresis represents a breakdown in the system of appropriate defecation, not through organic cause except through the mechanism of constipation. Normally, food is digested in the stomach and small intestine from which the waste products travel through the colon and shift into a semisolid state. When sufficient waste accumulates, colonic muscles contract, moving the waste toward the rectum, at the very distal end of the digestive tract. Stretching of the walls of the rectum creates sensation that leads to the urge to defecate. Usually at or before age 4 years, children learn to recognize this physiological urge and control it through voluntary contraction or relaxation of the external and internal sphincter muscles, leading to a bowel movement if the timing and place is appropriate, or through withholding when the time and/or place is not appropriate for evacuation (Fischel & Liebert, 2000). In the case of encopresis with constipation and overflow incontinence, the process of bowel evacuation becomes disrupted through repeated withholding of stool and leakage when impaction occurs. This can become a repetitive cycle that is not only dysfunctional but also painful and physically unhealthy for the child. When withholding occurs, moisture is reabsorbed into the body leaving a hard mass of stool that is increasingly difficult to pass. Additionally, fresh stool cannot be passed, but liquid can leak around the impaction causing overflow incontinence or staining. Some children also experience paradoxical constriction of the sphincter muscles when it is necessary and appropriate to have a bowel movement, further disrupting the process (Fischel & Liebert, 2000; Hales, Yudofksy, & Roberts, 2014). Over time with chronic constipation, the colon can become distended causing children to have reduced awareness or sensitivity to the physiological sensation indicating a need to defecate. They may also become desensitized to the odor from soiling (Ondersma, Ondersma, & Walker, 2001).

Associated Impairments and Comorbidities

Research has not been able to demonstrate a causal relationship whereby mental health conditions cause encopresis, with the exception perhaps of specific phobia related to toilets. However, it is thought that encopresis can lead to associated impairments in emotional and behavioral functioning during the course of the disorder (Kilanowski-Press, 2011) and that successfully treating the symptoms of encopresis can lead to other improvements in emotional and behavioral functioning (Young, Brennen, Baker, & Baker, 1995). For example, a group of boys diagnosed with encopresis between 8 and 10 years of age evidenced minimal mental and somatic disturbances 10 years later (Hultén, Jonsson, & Jonsson, 2005).

The impairments documented as comorbid with encopresis are numerous. Quality of life is one area that has been examined. Silverman et al. (2015) collected questionnaires from over 400 parents of children with encopresis and found that there were four common areas of health-related quality of life impairments among children with soiling and constipation. These included parental burden/distress, family conflict, worry about the social impact of encopresis and difficulties with the medical team treating the child. Other health-related concerns have been noted. Obesity prevalence was also found to be higher among children with functional constipation (Pashankar & Loening-Baucke, 2005), and urinary tract infections are also comorbid with encopresis, most often in females (APA, 2013).

Cox, Morris, Borowitz, and Sutphen (2002) also found that children with encopresis demonstrated psychological differences when compared with controls, although the severity of these symptoms remained in the subclinical range. These included significantly greater incidence of anxiety and depressive symptoms, attention problems, social problems, disruptive behavior, lower academic performance, and family environments characterized by less expression

and poor organization. Children presenting for treatment of encopresis tend to have a subclinical level of behavioral problems according to parent ratings on the Child Behavior Checklist (Achenbach & Edelbrock, 1983; Gabel, Hegedus, Wald, Chandra, & Chiponis, 1986). Encopresis can also adversely affect the child's self-esteem as a function of being socially ostracized by peers and treated negatively by parents because of their encopretic symptoms (APA, 2000).

Still, in some cases, there is evidence to support the existence of comorbid psychiatric conditions in the population of youth with encopresis. In fact, some studies have found psychiatric comorbidity in as high as 74% of children with encopresis with enuresis being the most common comorbid diagnosis. In a study by Unal and Pehlivantürk (2004), 30% of participants with encopresis also had a diagnosis of oppositional defiant disorder. Smaller cumulative percentages (less than 10% in each case) of comorbidity were also noted for other psychiatric conditions including attention deficit hyperactivity disorder, stereotypic movement disorder, mental retardation, anxiety disorder, conduct disorder, stuttering, depression, adjustment disorder, tic disorders, autism, and trichotillomania. Joinson, Heron, Butler, von Gontard, and Avon Longitudinal Study of Parents and Children Study Team (2006) examined a sample of more than 8000 children within the United Kingdom and found that children who soiled more than once per week had higher rates of bullying behavior (both as a victim and perpetrator), antisocial activities, attention and activity problems, obsessions and compulsion, and oppositional behavior. Encopresis may be more prominent in youth with ADHD (McKeown, Hisle-Gorman, Eide, Gorman, & Nylund, 2013) and specifically those with high hyperactivity scores (Johnston & Wright, 1993). Encopresis symptoms have also been found in higher rates among children with autism spectrum disorder with a family history of anxiety or obsessive-compulsive disorder (Peters et al., 2014).

It is these youth with clinical comorbidities who may experience the greatest difficulties responding well to treatment because of their psychiatric comorbidities. Yet, in some cases, encopresis has been effectively treated in children with comorbid emotional disturbance through combined medical and psychological approaches (Dawson, Griffith, & Boeke, 1990). In others, concurrent treatment of the psychiatric condition, ADHD, for example, has been shown to positively impact the symptoms of incontinence (Niemczyk, Equit, Hoffman, & von Gontard, 2015).

Course and Developmental Challenges

While it has been suggested that encopresis will spontaneously remit in many cases (Rockney, McQuade, Days, Linn, & Alario, 1996) the data to support this assertion has been limited (Rajindrajith, Devanarayana, & Benninga, 2013). The "wait and see" method has not been a recommended course of action because of the significant emotional distress and potential for physiological harm that can arise for the child during the course of the disorder (Mast & Smith, 2012; Mellon, 2012; Mikkelsen, 2014). Encopresis can intermittently exacerbate with time (APA, 2000), and a proportion of these cases continue into adulthood (Rajindrajith et al., 2013). Interestingly, however, encopresis is rarely diagnosed in individuals over 16 years of age (Kilanowski-Press, 2011). While this may be due to the large majority of cases being treated successfully within a year (assuming earlier diagnosis), an alternative explanation for this may be that the likelihood of referral to a clinician for problems with encopresis decreases with the age of the child. A 2005 population-based study in the Netherlands revealed that just over one-third of the 5- to 6-year-olds diagnosed with encopresis had seen a doctor to discuss their symptoms, and this decreased to just over one-fourth in the group of 11- to 12-year-olds with encopresis (van der Wal, Benninga, & Hirasing, 2005).

Diagnostic Procedures

Encopresis is a biobehavioral disorder, and thus evaluation for diagnosis should take into account both physiological and behavioral aspects that may contribute to the symptom presentation. Along with a comprehensive birth and developmental history, clinicians should obtain a detailed record of toilet training history and a history of stooling and dietary habits. Also relevant is an exploration of parent responses to encopresis symptoms (and those of others such as peers and teachers in the case of school-age children) and information surrounding environmental factors that were present at the apparent onset of the disorder (McKay & Storch, 2014).

Primary care physicians are often the first clinicians to examine children with encopretic behavior. They should assess for symptom characteristics (constipation, overflow incontinence, pain with bowel movements, quality of stool production, and anxiety about toileting), frequency of stools, and time spent in toilet sitting. Gastrointestinal imaging may be useful to determine the presence of gas or retained stool in the colon (APA, 2013). Lab work and other medical procedures, such as barium enema and anorectal manography (APA, 2013) and/or exam for colonic transit time (Koppen et al., 2016) may be needed to rule out organic causes (Mast & Smith, 2012). MacLean and Brayden (1992) reported a large number of medical conditions that may have encopresis-like symptoms as an associated feature. In these cases, an encopresis diagnosis would not be provided due to the organic etiology of those symptoms although it has been suggested that medical professionals may apply the diagnosis of encopresis more liberally (Kilanowski-Press, 2011). Possible organic causes for encopresis-like symptoms include Hirschsprung's disease, anal narrowing, anterior displacement of the anus, anal fistula, anal fissures, pelvic mass, hypothyroidism, hypercalcemia, meningomyelocele, cerebral palsy, irritable bowel disease, Crohn's disease, ulcerative colitis, lactose intolerance, pancreatic insufficiency, and use of medications such as narcotics, phenothiazines, tricyclic antidepressants, and iron supplements. In all, extensive medical testing is often unnecessary as organic causes (other than through the mechanism of constipation) account for only 5–10% of all cases of encopresis (Fischel & Liebert, 2000; Kilanowski-Press, 2011; Pashankar, 2005).

Prevalence rates are commonly noted at up to 3% of the pediatric population (over age 4 years per the diagnostic criteria) (APA, 2000; Coehlo, 2011) but may be as high as 7% worldwide (Hardy, 2009; Rajindrajith et al., 2013). Males outnumber females in diagnosis as much as 6:1 (Rajindrajith et al., 2013). Encopresis accounts for as much as 6% of psychiatric referrals among school-aged children (Hardy, 2009).

Treatment

Early treatment studies consisted mostly of single-case design but lent emerging evidence for the use of behavioral interventions in treating encopresis (Johnson & Van Bourgondien, 1977). Treatment for encopresis with or without constipation and overflow incontinence can be challenging and often met with frequent relapse. It often involves a multimodal approach that incorporates medical, educational, and behavioral interventions (Coehlo, 2011; Kilanowski-Press, 2011; Rajindrajith et al., 2013; Rowan-Legg & Canadian Paediatric Society, Community Paediatrics Committee, 2011) delivered by collaborating treatment providers in the fields of medicine and psychology (Howe & Walker, 1992). Although parents seeking treatment for their child are often overwhelmed amd frustrated and may even believe that their child's incontinence is intentional, family therapy absent medical and behavioral intervention is not usually effective (Ondersma et al., 2001), and parental readiness for treatment is an important factor in treatment outcomes (McKay & Storch, 2014).

The vast majority of the scientific literature reports very similar components to treatment plans for children with encopresis. Research suggests that there is a common, although perhaps not officially "standardized" treatment protocol that has proven to be effective (McKay &

Storch, 2014) although studies comparing two or more behavioral intervention protocols are virtually nonexistent (Freeman, Riley, Duke, & Fu, 2014). Behavioral strategies typically involve praise and reinforcement of independent toileting attempts and successful bowel movements in the toilet, toileting schedules, and having the child take charge of or assist with cleanup following soiling accidents (McKay & Storch, 2014). It is recommended that the behavioral treatment component also involve education for parents and children about encopresis, a non-accusatory approach from parents, and detailed record keeping of toileting behaviors (Koppen et al., 2016). Implementation of behavioral procedures across settings (such as at home and in school) is beneficial (Geroski & Rodgers, 1998).

Even absent a strong foundation of randomized controlled trials (RCTs) to document the effectiveness of a standardized behavioral protocol, combination interventions (behavioral and medical) appear effective for children with encopresis including those who have previously failed to respond to medical interventions alone (Ondersma et al., 2001). In 1976, Davis, Mitchell, and Marks evaluated the efficacy of a behavioral intervention program with 11 children diagnosed with encopresis. The program involved parents helping children to become aware of internal cues related to the need to defecate and implementing contingencies for appropriate toilet use. The authors reported that 7 of 11 participants were successfully treated and maintained progress at 7 months posttreatment follow-up. Many of these participants had previous poor response to physical and psychotherapeutic interventions. In a group of 110 children receiving counseling and education about encopresis, initial cleanout, and training in appropriate toileting methods, 1 year follow-up data supported significant improvements. Among this treatment group, 51% had no soiling accidents for over 6 months, and another 41% displayed at least some improvement in symptoms (Levine & Bakow, 1976). Lowery, Srour, Whitehead, and Schuster (1985) implemented a combined medical and habit-training program with 58 children diagnosed with encopresis. After initial cleanout, participants were trained to attempt toileting following specific meals and were made to use enemas after 2 successive days of not defecating. After 5 months, 60% of participants had achieved total continence, and another 23% had only staining. Frequency of incontinence decreased by 90% among participants not showing total symptom remission. At a 3-year follow-up, 51% of participants were still continent and only 10% had staining.

More recent research has continued to provide support for combined medical and behavioral intervention. Stark et al. (1997) demonstrated positive treatment response (absence of soiling by the end of treatment) in 86% of a participant group receiving only 6 total hours of combined intervention involving medical measures (enema clean-out and increased daily fiber intake) and behavioral measure (daily toilet sitting with differential attention, contingency management and contracting). Fireman and Koplewicz (1992) found that a significant decrease in soiling with evident in their 52 treatment participants within the first month of intervention and that nearly 85% of participants were accident-free for a period of 2 weeks or longer after approximately 1 month of treatment. Thus, behavioral intervention appears to be effective within a relatively short period of time. For those children who are resistant to parent-implemented behavioral interventions, adjunctive individual cognitive-behavioral therapy with the child may help boost treatment response rates (Knell & Moore, 1990).

Combined medical and behavioral interventions can be successfully delivered in a group format as well. Stark, Owens-Stively, Spirito, Lewis, and Guevremont (1990) educated families with previously treatment resistant children about enema use for cleanout, ways to increase dietary fiber, and appropriate toileting behaviors in small groups. They noted that soiling accidents were decreased by 83% from pre- to posttreatment. These gains were maintained and, in some cases, further improved 6 months following treatment completion.

The medical intervention component, particularly pharmacotherapy, may only be necessary in encopresis with constipation and overflow

incontinence (von Gontard, 2014). Treatment generally involves some form of bowel cleanout or removal of impacted feces to remedy a current state of constipation and prevent its recurrence medically until more appropriate toileting behaviors can be established. This cleanout involves use of a laxative or stool softener (Fischel & Liebert, 2000; Rowan-Legg & Canadian Paediatric Society, Community Paediatrics Committee, 2011).

Laxatives commonly used to treat encopresis include magnesium hydroxide, lactulose, liquid paraffin, and mineral oil. These have long-established use for fecal disimpaction in children (Pashankar, 2005) and have been shown to be effective and safe in combination both with increased fiber intake and through use of multiple laxatives concurrently (McClung et al., 1993). More recently, polyethylene glycol 3350 has been demonstrated as an effective evidence-based treatment that is safe for long-term use (Pashankar, 2005; Pashankar, Bishop, & Loening-Baucke, 2003; Pashankar, Loening-Baucke, & Bishop, 2003; Rowan-Legg & Canadian Paediatric Society, Community Paediatrics Committee, 2011). Loening-Baucke (2002b) demonstrated that polyethylene glycol 3350 was comparably effective to magnesium hydroxide over the course of 12 months in treating children with constipation and encopresis. Approximately two-thirds of participants in each treatment group were considered to be "doing well," although children receiving treatment with polyethylene glycol 3350 had significantly more soiling episodes. One-third of participants in the magnesium hydroxide group refused treatment, while none refused the polyethylene glycol 3350, making it an acceptable alternative laxative treatment for children with encopresis. Polyethylene glycol 3350 has also been shown to be as effective as liquid paraffin in treating symptoms of constipation, with less adverse side effects (Rafati, Karami, Salehifar, & Karimzadeh, 2011). After initial cleanout, laxatives can be and are used long-term as part of a maintenance program, with the goal being that children have up to 3 stools per day without soiling (Mast & Smith, 2012).

Biofeedback has also been explored as a possible adjunctive treatment approach for encopresis. Initial study suggested that a treatment protocol combining biofeedback with medical and behavioral intervention (referred to as enhanced toilet training) was as effective as combined medical and behavioral intervention when compared against treatment using laxatives alone (Cox, Sutphen, Ling, Quillian, & Borowitz, 1996). Borowitz, Cox, Sutphen, and Kovatchev (2002) later compared the effects of three different levels of treatment for 87 children with encopresis who had at least weekly soiling episodes for 6 months or more. They compared outcomes in a group receiving intensive medical treatment only, a group receiving intensive medical treatment plus behavioral intervention (enhanced toilet training), and a group receiving intensive medical treatment, enhanced toilet training and biofeedback. At 1 year posttreatment, the cure rates (defined as a child having less than 1 soiling episode in a 2-week period) were not significantly different between the treatment groups. However, consistent with earlier research, symptom improvement rates (defined as a reduction in soiling) were observed to be greatest among the group treated with intensive medical treatment plus enhanced toilet training. These results suggest that biofeedback was not a necessary addition to treatment protocols.

Other literature including RCTs (van der Plas, Benninga, Redekop, Taminiau, & Büller, 1996) and meta-analysis have also failed to find significant benefit of biofeedback training for encopresis that is not due to organic causes (Brazzelli, Griffiths, Cody, & Tappin, 2011). Thus, although it may be useful for constipation resulting from physiological causes such as pelvic floor dysfunction (Croffie et al., 2005), it does not appear to be a necessary or sufficient treatment method for functional encopresis with constipation and overflow incontinence (Mellon, 2012). One of the only studies to demonstrate superiority of biofeedback as compared with medical intervention, via laxative treatment, was conducted with children having nonretentive encopresis (encopresis without constipation

or overflow incontinence). In this case, biofeedback alone produced significantly high success rates, whereas laxative therapy appeared to have no added benefit to the children receiving treatment (van Ginkel et al., 2000). This is not entirely surprising given that children with the nonretentive form of encopresis do not require bowel cleanout.

A more recent development in the encopresis treatment literature has been the emergence of internet-delivered behavioral interventions. Programs have been created relatively recently and continue to undergo empirical validation. If evidence consistently supports the efficacy of these interventions, it may have a substantial impact on the prevalence of the disorder. It has been suggested that due to the nature of encopresis, parents may be hesitant to seek out professional help, and thus prevalence rates may underestimate the commonality of the condition (van der Wal et al., 2005).

Home-based therapy has been touted as a potentially useful alternative to other outpatient treatment models (Nolan & Oberklaid, 1993). Ritterband et al. (2003) compared to the added benefits of internet-delivered behavioral intervention (enhanced toilet training) to a standard treatment protocol delivered in a physician's office. They found that the participants receiving internet-based intervention demonstrated significantly increased rates of defecation in the toilet, increased unprompted toilet visits, and reduced fecal soiling. Web-based interventions have been shown to be effective in reducing soiling accident frequency by a minimum of 50% in almost all of children participating in the intervention (95%) (Ritterband et al., 2008). An initial evaluation of a fully automated internet-based intervention called UCanPoopToo revealed that children with encopresis who completed the intervention program had significantly fewer soiling accidents both at the initial follow-up (4–6 weeks) and 1-year follow-up when compared to those receiving standard care alone. Impressively, participants in this study had an almost 4-year history of bowel accidents and were able to decrease the frequency of those accidents by as much as 50% (Ritterband et al., 2013).

Prognosis and Outcomes

Treatment of encopresis is successful for a substantial portion of those treated, but it may take several months to years before symptoms remit completely, and relapse rates are substantial even among those with initially positive treatment response (Catto-Smith, 2005; Kilanowski-Press, 2011; Pashankar, 2005; van Ginkel et al., 2003). van Ginkel et al. (2003) noted higher relapse rates among males. Recovery rates at 1 year following diagnosis have been reported between 30% and 50%, increasing in some cases to between 48% and 75% after 5 years (Loening-Baucke, 2002a). Patel and Pratt (1999) reported that 50–60% of children achieve acceptable bowel control (defined as an absence of soiling) within 1 year. Others have reported limited outcomes, with only one-third of those treated showing complete recovery after 3-and-a-half years despite reporting they found treatment methods to be acceptable and helpful (Bernard-Bonnin, Haley, Bélanger, & Nadeau, 1993). Factors associated with relapse include early onset of symptoms (prior to age 4 years), and the subtype of encopresis involving constipation with overflow incontinence (Catto-Smith, 2005). Among children with the overflow incontinence subtype of encopresis, those whose symptoms begin prior to achieving fecal continence may be at the greatest risk for poor treatment response (Taubman & Buzby, 1997). Others have found nearly the opposite, whereby children who are treatment resistant tend to have less stool withholding upon initiating treatment but had accidents at all hours of the day (Landman, Levine, & Rappaport, 1984).

Rockney et al. (1996) conducted a long-term follow-up with children diagnosed and treated for encopresis. Of the 45 participants retained beyond 1 year for follow-up (average follow-up was 53 months), 58% remained in remission and 29% continued with improvement in symptoms. Predictors of positive outcome were diagnosis of constipation with overflow incontinence subtype and an absence of previous treatment.

Unal and Pehlivantürk (2005) examined the long-term clinical outcome in children treated for

encopresis. Six years after treatment, approximately 84% of the children treated continued to evidence a complete recovery from symptoms. Factors predictive of positive outcome included an absence of constipation, and diagnosis within 1 year of symptom onset. Participants in this study who responded best to treatment also displayed stronger academic performance and higher parental education level when compared with treatment-resistant counterparts.

Other research has demonstrated that social or environmental factors are correlated with prognosis. For example, Alpaslan, Koçak, Avci, and Güzel (2016) noted that children with encopresis displayed worse treatment outcomes if their parents were of low education level, low socioeconomic status, or if their family was characterized as divorced, disorganized, or chaotic. Among children with encopresis, families are also characterized by more strictness and less psychological autonomy (Akdemir, Çengel-Kültür, Saltik-Temizel, & Senses-Dinç, 2015; Çengel-Kültür, Akdemir, & Saltik-Temizel, 2014). Kammacher Guerreiro, Bettinville, and Herzog (2014) found that longer disease duration, male gender, and a history of stool retention were all predictors of poor prognosis. Poor prognosis has also been documented in cases where children with behavioral problems prior to treatment, particularly oppositional behavior, have one or both parents who possess a negative attitude about their child's encopresis symptoms. This attitude, often characterized by belief that the child's behavior is intentional and can be willingly controlled if the child desired, can interfere with consistent implementation of behavioral contingencies for appropriate toileting behavior and undermine treatment success (Young, Brennan, Baker & Baker, 1996).

Research also exists that demonstrates a correlation between family characteristics and an increased risk of encopresis diagnosis. For example, children with the constipation and overflow incontinence subtype of encopresis are more likely to have a mother with psychiatric symptoms (Akdemir et al., 2015; Çengel-Kültür et al., 2014). Joinson et al. (2008) observed that maternal depression and anxiety, as well as developmental delays and difficult temperament, positively predicted later diagnosis of encopresis among a sample of more than 10,000 school-aged children in the United Kingdom.

It has been suggested that adolescents with chronic encopresis require a different treatment approach because of their developmental stage and difficult family dynamics that are commonly present in such cases (Fennig & Fennig, 1999). Fennig and Fennig demonstrated, using four case studies, that adolescents with chronic encopresis had positive response to an integrative treatment approach involving inpatient care in a medical-psychiatric unit. Adolescents were in treatment for an average of 2 weeks receiving combined medical and behavioral intervention. Parents also received education and guidance. This approach resulted in complete remission of symptoms in the four case studies presented. More recently, however, a small intervention study with two adolescents with encopresis and comorbid autism spectrum disorder and intellectual disability demonstrated efficacy of a combined medical and behavioral approach implemented across home and school settings with both participants achieving full bowel continence within 10 weeks and maintaining treatment gains even after laxative use was discontinued (Axelrod, Tornehl, & Fontanini-Axelrod, 2016). This lends support to the notion that treatment protocols need not vary from those proven successful with younger children. Even among youth initially treated during childhood, symptoms of constipation may persist into adolescence. van Ginkel et al. (2003) followed 418 children (aged 5 years or older) with constipation for up to 8 years post-diagnosis and found that despite positive initial response to treatment, 30% of participants aged 16 and older still had problems with constipation.

Treatment options for non-retentive encopresis (encopresis without constipation or overflow incontinence) are sparse due to it being the less common subtype. Thus less is known about positive predictors and risk factors related to treatment outcomes. It has been noted that 15% of children with this subtype of encopresis still

demonstrate the same symptoms at age 18 years (Rajindrajith et al., 2013) while others have noted that children with the non-retentive form of encopresis fare better compared with children having the constipation and overflow incontinence subtype of encopresis (Mohammed & Mekael, 2012).

Conclusions

Encopresis is a biobehavioral disorder characterized by the repeated passage of fecal matter in inappropriate places either involuntarily or intentionally. It is diagnosed only in individuals over the developmental age of 4 years who display symptoms without identifiable organic etiology, except as a function of constipation. Despite being included in the DSM since its second edition, relatively limited research exists around its etiology and efficacious treatment approaches. Particularly lacking are RCTs that compare standardized treatment protocols against one another. However, it is common practice to implement combined approaches using medical, educational, and behavioral methods to teach families about the disorder and its symptoms, to improve diet for aiding in ease of defecation and/or using medicinal treatment to clear out bowels and training families in behavioral strategies to develop more positive and successful toileting habits. Encopresis is noted in up to 8% of the pediatric population and is far more common in males. The subtype involving constipation and overflow incontinence is more common, and more research exists with participants having this subtype of the disorder. Numerous factors have been identified as predicting poorer prognosis and outcomes within this population including male gender, chronicity of symptoms, earlier symptom onset, comorbid behavioral disorders, and lack of parental cooperation or other familial factors that may lead to difficult complying with treatment regimens. This includes factors like single-parent, large, or otherwise chaotic families, parental mental illness, and negative parent attitudes about encopresis to name a few.

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Part XI

Externalizing Disorders: Disruptive, Impulse Control and Conduct Disorders

Oppositional Defiant Disorder

25

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Ryan was a 15-year-old male referred for therapy by his mother because of significant behavior problems he had been having at home and school. During the intake interview, his mom expressed frustration that just yesterday he had ripped out all of her newly planted roses, as he was upset that she would not drive him to go to a friend's house after school. Ryan's mother reported that he would not listen to anything she said at home anymore, did not complete chores, would come and go from the house as he pleases, and argued and became upset anytime he was told "no" or not allowed to do something. When he would become really upset with his mother, he would do things such as hide her car keys or cell phone. Ryan had never run away from home, but he frequently left without permission and would not come home for several hours. At the time of the intake, Ryan had never been arrested; however, his mother was concerned about the possibility of this as he had friends who had been caught shoplifting.

Ryan's mother stated that Ryan had always been a child to ask "why" and question rules, but this behavior had increased to a degree that he now would negotiate or argue even simple requests. She reported that it was in the fifth grade when she started having significant problems at home, and in the seventh grade when his argumentative behavior escalated to the point that he began getting in more trouble at school and was even suspended. He had been an avid basketball and soccer player when he was younger, and now he no longer wanted to participate on any teams. His refusal to play sports also coincided to a change in his friend group. His mom did not like his new friends, claiming they were bad influences and he was acting in ways "he never would otherwise." While he had not gotten in any fights at school, Ryan had received several referrals for talking back to teachers, refusing to follow classroom rules, and even arguing with the principal when in trouble. His mother described his typical mood as "crabby" and said he loses his temper several times a day. He was not physically aggressive at home, but would yell and slam his door when upset. In the eighth grade, Ryan participated in a 6-week group anger management class, and he had subsequently seen two different therapists for individual counseling. He saw one counselor for only three to four sessions, and the other he saw weekly for 6 months. Ryan described these experiences as "a waste of time," and his mom admitted that there had been little improvement in his behavior despite participating in individual therapy.

After meeting with Ryan and his mother, it did not appear that there were any major life changes that may have contributed to the change in Ryan's

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behavior in the fifth grade. He did not present with symptoms of depression or any other internalizing disorders, and there were no significant social communication deficits or other identifiable cognitive impairments that would explain his difficult behavior at home and school. Rather, it appeared that Ryan met the criteria for oppositional defiant disorder (ODD). This diagnosis is characterized by the American Psychiatric Association [American Psychiatric Association (APA), 2013] as a persistent pattern of negative, hostile, defiant, and disobedient behaviors toward others. The frustrations and difficulties experienced by Ryan's mother are similar to those experienced by many other parents and adults working with children and adolescents with ODD. Parents and teachers commonly complain that these adolescents "feel like the rules do not apply to them" or "just do what they please" regardless of societal norms, rules, or adult requests. Any individual or professional who works with children and adolescents can attest to the fact that defiance, rule breaking, and arguing are common and are even developmentally appropriate, in many children and adolescents. But when does "typical" teenage defiance become developmentally *inappropriate*? What constitutes a level of defiance significant enough for one to be provided a mental health label? Or specifically, at what point do defiant and oppositional behaviors qualify one for the diagnosis of oppositional defiant disorder? Of more importance, however, may be answers to the question of how one goes about treating or providing interventions for significant defiance, as the problems experienced by Ryan's mother with poor treatment effectiveness are very common. These questions have been debated since the diagnosis of ODD was included in the DSM, and researchers are still working to identify the most appropriate treatments for this disorder.

The purpose of the present chapter is to provide background on ODD and the presentation of symptoms needed for one to qualify for a diagnosis under the *Diagnostic and Statistical Manual of Mental Disorders – Fifth Edition* (DSM-5). In addition, this chapter will provide information on the etiology of the disorder, developmental progression and presentation, and comorbid conditions that often exist in children and adolescents with ODD. The difficulties and impairments often observed will be discussed, as well as issues related to the treatment of ODD.

Diagnosing Oppositional Defiant Disorder

While the American Psychiatric Association first introduced the DSM in 1952 to help professionals more accurately diagnose mental health disorders, childhood and adolescent disorders related to disruptive and delinquent behaviors (i.e., conduct disorder and oppositional defiant disorder) were not included until the second edition of the DSM (APA, 1968). The DSM-II provided two categories of disruptive behavior disorders, including *group delinquent reaction of childhood* and *unsocialized aggressive reaction of childhood.* Both of these disorders were largely related to delinquent and antisocial youth. *Group delinquent reaction of childhood* related to those adolescents identifying with the behaviors and values of a delinquent peer group or gang, while *unsocialized aggressive reaction of childhood* was a diagnosis for children and adolescents who engaged in hostile, disobedient, aggressive, and other antisocial acts (e.g., lying, bullying, fighting). It was not until the DSM-III that a disruptive behavior was added to account for children and adolescents with extreme defiant behavior, at which time it was labeled *oppositional disorder* (APA, 1980). To qualify for a diagnosis of oppositional disorder, a youth needed to display at least two defiant or oppositional symptoms over a 6-month period, with diagnostic symptoms including violation of minor rules, temper tantrums, argumentativeness, provocative behavior, stubbornness, and even swearing.

The inclusion of oppositional disorder in the DSM-III was progress in more accurately identifying, diagnosing, and treating children and adolescents with disruptive behaviors that interfered with their functioning; however, the diagnostic criteria for this disorder were rather undefined and largely controversial. Even with slight

revisions in diagnostic criteria for the DSM-III-R (APA, 1987), the reliability and validity of the diagnosis was debated, and they have continued to be debated throughout subsequent revisions of the DSM (Pardini, Frick, & Moffitt, 2010). Researchers and professionals alike have criticized the overlap between symptoms of oppositional disorder and conduct disorder (CD), another disruptive behavior disorder characterized by a more severe violation of the rights of others (e.g., stealing, assault). In addition, the lack of specificity of symptoms (e.g., how to qualify extreme stubbornness) has been critiqued, as well as the limited empirical evidence supporting the reliability of the diagnostic symptoms of ODD (especially in populations outside of delinquents), the limited research examining ODD in females, and the possible over-pathology of typical childhood or adolescent behaviors (e.g., Angold & Costello, 1996; Lahey, Loeber, Quay, Frick, & Grimm, 1992; Nock, Kazdin, Hiripi, & Kessler, 2007; Rey et al., 1988; Russo, Loeber, Lahey, & Keenan, 1994). Because of these ongoing criticisms related to accurately diagnosing ODD, various workgroups and professional groups have formed over time to help clarify diagnostic criteria with each revision of the DSM as well as to provide empirical support for continuing to include the diagnosis in the most current edition (DSM-5). With each subsequent revision of the DSM there has been an attempt to provide more specific diagnostic criteria, address the limited research related to ODD, identify potential subtypes of ODD, and to better address difficulties accurately differentiating ODD from other disruptive behavior disorders such as CD (Herzhoo & Tackett, 2015; Leadbeater & Homel, 2015; Pardini et al., 2010). Nevertheless, there does remain some degree of subjectivity in what is considered pathological defiance versus what is developmentally appropriate.

DSM-5 Diagnostic Criteria for ODD

The DSM-5 places ODD in the category of disruptive, impulse control, and conduct disorder, all disorders which are directly related to both (a) one having difficulty controlling their emotions or behaviors *and* (b) poor self-control resulting in a direct violation of the rights of others and/or resulting in conflict with societal norms or the legal system (APA, 2013). In regard to current diagnostic criteria, the DSM-5 specifies that to qualify for a diagnosis of ODD, one must meet several different criteria, as described below.

(a) *Display a pattern of an angry/irritable mood, argumentative/defiant behavior, or vindictive behavior in interaction with others*

The DSM-5 provides several examples of behaviors that would demonstrate this type of criteria, including: frequently losing one's temper, being easily annoyed, often being angry or resentful, often arguing with adults or authority figures, actively refusing to comply with requests or rules from authority figures, deliberatively annoying others, blaming others for one's mistakes or misbehavior, or being spiteful or vindictive. In order to meet the criteria for demonstrating a *pattern* of behavior, there must be at least four of these aforementioned behaviors present for at least a 6-month period of time. In the case above of Ryan, his pattern of defiant/disruptive pattern of behavior was evidenced by the fact that for approximately 2 years he had demonstrated chronic irritability, regularity of losing his temper, arguing frequently with adults both at home and school, often refusing to follow requests or rules, and being spiteful or vindictive by doing things such as hiding his mother's keys out of anger or tearing out her roses.

(b) *Experience significant impairment of functioning as a result of these behaviors*

This impairment may occur in one's social, educational, or occupational functioning, and/or it may impair relationships by causing significant distress to significant others. Social impairments may be observed in an increase in negative peer relationships (e.g., hanging out with friends engaging in delinquent behavior), a loss of friendships, or a youth no longer engaging in social activities. Impairment causing distress to

others may be evident at home by increased fighting and hostility among family members, broken familial relationships, or even indirectly with parents having employment difficulties after needing to miss work to deal with defiant-related behaviors (e.g., attending school discipline meetings). In regard to school impairments, the defiance may result in ongoing school suspension or discipline, which may lead to poor grades, delayed graduation, or even school dropout. In Ryan's case, his behaviors were contributing to poor school performance (impairment on educational functioning), he was no longer participating in sports or group activities (impairment on social functioning), there was a stressful relationship with his mother (impairment on social functioning), and his mother was experiencing significant stress related to managing Ryan's difficult behavior and worrying about the consequences it was having (impairment causing distress to significant others).

(c) *Display a pattern of behavior does not occur exclusively during the course of another disorder*

Specifically, the pattern of defiant behavior cannot occur only in the midst of an episode related to bipolar or disruptive mood dysregulation disorder, substance use, or depressive disorder. If a child had been diagnosed with bipolar disorder, for example, and the defiant behaviors occurred in the context of a depressive or hypomanic episode, then ODD would *not* also be diagnosed as the defiance could be attributed to bipolar disorder. It is important to rule out another disorder as the major cause of the defiant and oppositional behavior, as some disorders have externalizing behaviors associated with them. For example, although depression is commonly associated with symptoms like sadness and withdrawal, disruptive behaviors such as irritability and oppositional defiance are frequently observed in children with depression (APA, 2013; Jacobs et al., 2010). It is also not uncommon for youth abusing substances to be more irritable, less compliant with rules, and more argumentative. However, if these behaviors appear to be directly related to withdrawal from substances, an attempt to obtain substances, or behaviors that happen while under the influence, then ODD would not be diagnosed in addition to the substance use disorder. While Ryan did present with some indication of social withdrawal, there were no other symptoms to indicate he was experiencing depression, and there was no history of substance use. After a comprehensive psychological evaluation, it was determined that his oppositional and defiant behaviors did not appear to be related to any other mental health disorder, but rather that his behaviors appeared to be directly the result of ODD.

Comorbidity While it is important to rule out mood and substance use disorders as primary contributing factors to oppositional and defiant behaviors, this does not imply that ODD does not commonly occur with other mental health disorders. Rather, ODD is often found to be comorbid with other mental health disorders. Nock and colleagues (2007) conducted a nationwide survey of over 9000 participants and found that 92.4% of them qualified at some point for another mental health disorder. The lowest rates of comorbidity with ODD were anxiety disorders, whereas the highest rates of comorbidity were other impulse control disorders. This is consistent with other studies in that ODD has most commonly been associated as being comorbid with other disruptive behavior disorders such as attention-deficit/hyperactivity disorder (ADHD) and CD. It has been suggested that upward of 50% of youth with ADHD or CD will also qualify for a diagnosis of ODD (APA, 2013), indicating a very high comorbidity between these disorders. Interestingly, many practitioners consider a diagnosis of ODD a precursor to CD, even though the many youth diagnosed with ODD will never develop CD (Nock et al., 2007).

Background of ODD

Etiology

Similar to most other mental health disorders, there is no one specific cause for ODD that has been identified. Rather, research has suggested a variety of factors that may contribute to the

development of disruptive behaviors such as ODD. For example, a number of studies have linked prenatal substance exposure with disruptive behaviors (e.g., Badu et al., 2007; Linares et al., 2006; Russell et al., 2015; Spears, Stein, & Koniak-Griffin, 2010), as well as a variety of environmental factors such as exposure to violence, living in high-crime neighborhoods, low socioeconomic status, and minority status (e.g., Eiden, Coles, Schuetze, & Colder, 2014; Vanfossen, Brown, Kellam, Sokoloff, & Doering, 2010, White & Renk, 2012). In addition to the likelihood of there being multiple factors contributing to the development of ODD, when discussing research related to the etiology of ODD, it is important to note that overall there is limited research examining etiological factors specific to ODD. Rather, the majority of available research focuses on the common risk factors associated with all of the disruptive behavior disorders and often combines symptoms together of ODD and CD (APA, 2013; Burke, Loeber, & Birmaher, 2002). In addition, often times, research uses samples of youth with more severe disruptive behaviors, such as samples of juvenile delinquents. Despite this need for more focused research on the risk factors associated with ODD, studies have generally found common etiological influences, including both biological and environmental influence (Bornovalova, Blazei, Malone, McGue, & Iacono, 2013; Burke et al., 2002; Martel, Nikolas, Jernigan, Friderici, & Nigg, 2012).

Though evidence is limited, there is some research to suggest that there may be biological influences on the development of ODD. It has been proposed that a child's temperament, such as his/her inflexibility or intense and reactive way of responding, may be associated with the development of ODD (Burke et al., 2002). Adolescents with ODD have also been found to have lower heart rates and skin conductance activity, reduced basal cortisol reactivity, and abnormalities in the prefrontal cortex and amygdala (Burke et al., 2002). The prefrontal cortex and amygdala are two areas of the brain associated with emotion processing and behavioral/emotional regulation, and consistent with the findings by Burke and colleagues (2002), others have also found evidence that disruptive behavior disorders such as ODD are likely related, in part, to functional differences in these areas of the brain (Beauchaine, Gatzke-Kopp, & Mead, 2007; Bellani, Garzitto, & Brambilla, 2010).

Martel and colleagues (2012) also found evidence for biological markers as a pathway to the development of ODD. These researchers examined a sample of over 500 children, 148 of whom were diagnosed with ODD, and found that those with ODD had differences in neurotransmitter functioning. Specifically, they found dysfunction with the neurotransmitter serotonin, which is important in mood regulation. These researchers argued that this dysfunction is likely a contributing factor to emotional dysregulation, which can subsequently contribute to disruptive and defiant behaviors for youth who reside in a high conflict family environment. Essentially, while biological markers were noted, these researchers argued that ODD is caused by an interaction of biological, familial, and environmental factors.

While it is difficult to tease out the interaction between genetics and environment, research has suggested that youth are more likely to develop disruptive behavior disorders such as ODD if one or both of their parents have antisocial behavioral tendencies. Bornovalova and colleagues (2013) conducted a study of 1255 families with same-sex twin pairs aged 11 years old to determine the effects of parental antisociality, ineffective parenting practices, and marital problems on the development of disruptive behavior disorders (i.e., ADHD, ODD, and CD). Results indicated that youth whose parent/s had antisocial tendencies scored higher on symptoms of disruptive behaviors such as defiance.

In terms of environmental factors, studies have found that maladaptive parenting practices and high conflict family environments put youth at risk for the development of ODD. In particular, maladaptive parenting practices such as parents who use more punitive discipline strategies, who lack warmth or involvement in their child's life, or who have a higher number of interpersonal conflicts with their children are shown to put youth at higher risk of ODD (Bornovalova et al., 2013; Burke et al., 2002; Martel et al., 2012). Moreover, some evidence suggests that disruptive behavior

disorders are more common in children whose parents are divorced, experience marital discord, or who come from lower socioeconomic homes (Bornovalova et al., 2013; Loeber, Burke, Lahey, Winters, & Zera, 2000).

There are several theories that take these various factors into consideration to explain the development of disruptive behavior disorders such as ODD. One such theory is the *hostile attribution bias*, which relies on the notion that individuals with disruptive behavior disorders are more likely to misperceive the actions and intentions of others as hostile or threatening, and in turn the adolescent responds to a situation in a negative way (APA, 2013, Dodge, Price, Bachorowski, & Newman, 1990). Another theory that attempts to explain the multifaceted nature of disruptive behaviors and conduct problems is the *social information processing model* (Crick & Dodge, 1994; Lemerise & Arsenio, 2000). This model states that it is a combination of cognitive distortions (such as hostile attributions) and impaired cognitive processes that result in poor social interactions and negative relationships and interactions with others. The *coercive parent-child interaction theory,* on the other hand, argues that a major contributing factor to escalated disruptive behaviors is poor communication and exchanges between parent and child as well as inconsistent discipline practices that result in more negative interactions and a cyclical pattern of increased communication and parenting difficulties (Patterson, 1982, 2002). These theories are also used to help explain disruptive behaviors associated with CD (APA, 2013).

Developmental Progression

The prevalence of ODD has been estimated to range from 2% to 16% of children and adolescents (APA, 2013). Similar to other disruptive behavior disorders, ODD is more common in males than females. According to the DSM-5, symptoms of ODD are often first evident in early childhood, with behaviors progressing through and peaking during adolescence. In a study conducted by Nock et al. (2007), the onset for ODD was found to range from 7 to 13 years old, with a median age of onset at 12 years old. The study by Nick and colleagues (2007) found results similar to other studies in that, while the duration of symptoms for ODD vary between adolescents, the duration is often negatively correlated with earlier age of onset and positively correlated with symptoms severity. Specifically, those who develop ODD at a younger age are likely to display symptoms for a longer period of time as well as symptoms that are more severe in nature. Consistent with this, Burke and colleagues (2010) found that youth who met criteria for ODD at the age of 4–6 years old continued to have significant functional impairments throughout adolescence.

It is often assumed that ODD is merely a precursor to CD or other antisocial behaviors; however, this is typically not the case. Although they may continue to experience difficulties in social functioning or other impairments, most children or adolescents who are diagnosed with ODD do not go on to develop a more severe disruptive behavior disorder (Nick et al., 2007; Rowe, Costello, Angold, Copeland, & Maughan, 2010). That being said, many of those who are diagnosed with CD previously qualified for a diagnosis of ODD. Related to this, several developmental pathways have been suggested in the development of disruptive behavior disorders. For example, the *authority conflict pathway* asserts that disruptive behaviors begin prior age 12, when youth begin displaying stubborn and defiant behavior, which progress to defiance and authority avoidance (e.g., truancy and running away). The *overt pathway* suggests that youth begin displaying minor aggressive and defiant behaviors, with this later progressing to more significant defiance, physical fighting, and violence. The *covert pathway* suggests that symptoms typically appear before the age of 15, begin with minor covert behaviors (e.g., defiance, rule breaking), and progress to property damage (e.g., fire setting and vandalism) and moderate to serious delinquency (Burke et al., 2002). Similarly, while supporting research remains limited, a hierarchical pathway between ODD, CD, and antisocial personality disorder (APD) has also been suggested (APA, 1994; Burke, Waldman, & Lahey, 2010). This pathway is built on the premise that although the majority of youth with ODD will

never meet criteria for CD, youth with ODD are at a four times greater risk of developing childhood-onset CD than youth in the general population. Of those with childhood-onset CD, approximately one-third will meet the criteria for a diagnosis of APD in adulthood (Burke et al., 2010). Essentially, these various developmental pathways suggest that, while defiance does not indicate one has a high chance of later behavior difficulties, those who display more severe antisocial behaviors may have first displayed defiance and oppositional behaviors.

In terms of prognosis, the literature suggests that, while youth with ODD can experience many social, academic, and familial difficulties (as discussed below), the specific symptoms of defiance typically do not progress into adulthood. When compared to youth with CD, those with ODD typically have a much better prognosis (Burke et al., 2010). For example, youth diagnosed with ODD tend to have lower rates of school-related difficulties (e.g., suspensions) and contact with law enforcement (Loeber et al., 2000) than youth with CD. That being said, there is research to suggest that, while youth with ODD often show considerable improvement in symptoms and functioning, some may experience functional difficulties across the life-span (Burke et al., 2010). Ferguson, Boden, and Harwood (2010) found that individuals with ODD had a greater risk of poor employment outcomes into adulthood. The authors also found that ODD in adolescents is associated with a greater long-term risk internalizing problems, such as depression, anxiety, and suicide attempts. Burke, Rowe, and Boylan (2014) also found that, while the specific symptoms of ODD (i.e., irritability and defiant rule-breaking behavior) may wane over time, it is not uncommon for those with ODD to have more social and employment difficulties in early adulthood.

Risk Factors and Negative Implications Associated with ODD

ODD has been associated with a variety of risk factors and difficulties, including impairments in cognitive and academic performance, as well as family and social functioning.

Historically, research has been inconsistent regarding whether there is a relationship between ODD and low average intelligence (Frazier, Demaree, & Youngstrom, 2004; Lynham & Henry, 2001). However, research has long found an association between language difficulties and disruptive behaviors (e.g., Silva, Williams, & McGee, 1987; St. Clair, Pickles, Durkin, & Conti-Ramsden, 2011; Stevenson, McCann, Watkin, Worsfold, & Kennedy, 2010; Yew & O'Kearney, 2012; Conti-Ramsden, 2013). In addition, research over the past decade has increasingly supported a link between disruptive behaviors and executive functioning deficits. Executive functions are those cognitive abilities related to self-regulation, and they include things such as attention and concentration, impulse control, organization, planning, and flexible thinking. A recent meta-analysis by Schoemaker, Mulder, Devokic, and Matthys (2013) specifically found that children with ODD showed executive functioning deficits in working memory, impulse control, and flexible thinking. These deficits have been significant enough that some have argued that this may explain the high comorbidity between disruptive behavior disorders like ODD and ADHD (Thorell & Wahlsted, 2006).

Another significant risk factor associated with ODD is academic difficulties or poor school performance. While the deficits experienced by youth with ODD are often not as significant as those with CD, students who have been diagnosed with ODD are at a greater risk of having reading difficulties and may display poor academic performance in reading, writing, and math. While many of these academic difficulties may be related to the high comorbidity between ODD and ADHD (as academic difficulties are highly associated with ADHD), researchers have found that even independent of ADHD youth with ODD struggle academically (Sayal, Washbrook, & Propper, 2015). There is also a higher likelihood of these youth receiving special education services, with one study finding that nearly half of students receiving special education services for behavior-related difficulties met the criteria for ODD (Déry, Toupin, Pauzé, & Verlaan, 2004). Not surprisingly, children and adolescents with ODD are also more likely to have a history of

truancy, suspensions, expulsions, and school dropout. This is typically a direct implication of their defiance, as inherent in ODD symptoms is an unwillingness to follow school rules and tendency to argue frequently with adults. Students with ODD may be more inclined to challenge classroom rules, refuse to complete in class tasks, fail to complete homework, or have a tendency to "test the limits" with teachers and/or argue with their peers (Sayal et al., 2015). These types of behaviors in the school environment can cause significant difficulties that result in detention, in-school suspension, out-of-school suspension, or even expulsion if the disruptive behaviors are frequent or severe enough.

Also of significant note is the fact that ODD is highly correlated with social difficulties. Many of the symptoms of ODD – irritability, argumentativeness, and anger – are likely to cause strain in the relationships that these youth have with family, friends, or others in the community (APA, 2013; Burke, Pardini, & Loeber, 2008). In the family environment, research shows that adolescents with ODD tend to have more familial dysfunction and hostility, with parents being more likely to use inconsistent discipline, have more negative interactions with their child, and increase stress and mental health difficulties (Duncombe, Havighurst, Holland, & Frankling, 2012; Greene et al., 2002).

Children and adolescents with ODD tend to have a greater number of poor peer interactions, a tendency to navigate toward negative peer influences, and are more likely to experience rejection from their peers (Munkvold, Lundervold, & Manger, 2011; van Lierr & Koot, 2010). A study that followed long-term implications of ODD even found that many of these social difficulties persist into early adulthood (Burke et al., 2014). These researchers followed a sample of 177 males who were diagnosed with ODD between ages 7 and 12 and found that in early adulthood these youth had poorer peer relationships, had more difficulties developing and sustaining romantic relationships, and often had ongoing strain in their relationships with parents. In addition, those with a history of ODD had employment difficulties. For example, the study found that these individuals were more likely to not have appropriate references for job applications.

Youth with ODD are also more likely to have legal difficulties and become involved in the juvenile justice system than their typically developing peers (Pardini & Fite, 2010), and many delinquents are diagnosed with ODD. Findings from a large study found that 40% of the youth in the juvenile justice system met criteria for ODD (Mericle, McClelland, Abram, Teplin, & Dulcan, 2002). In comparison to the prevalence of 2% to 16% in the general population, this is an astonishingly high rate of ODD in the juvenile justice system. Delinquents with ODD are more likely to have ongoing involvement with the juvenile justice system (Pardini & Fite 2010), and researchers have found that ODD symptoms were associated with increased levels of conduct problems and a greater chance of being charged for serious crimes. More specifically, Ferguson, Boden, and Harwood (2010) further found that ODD is related to property and violent offenses, arrests, and overall increased risk of crime. This level of seriousness in behavior is not required for a diagnosis of ODD, and often times those behaviors that would lead to legal involvement may indicate a youth is displaying symptoms more consistent with CD. Nevertheless, the tendency toward rule breaking behavior, noncompliance, and arguing with adults puts children and adolescents at a greater risk of legal involvement (APA, 2013). Gang-affiliated juveniles, for example, show higher rates of ODD when compared to non-gang-affiliated juveniles (Sargent et al., 2013). Gang affiliation come with its own set of risk factors, such as typically displaying more delinquent behavior, committing more crimes, and having higher rates of substance abuse (Klein & Maxson, 2006; Sargent et al., 2013).

Treatment of Oppositional Defiant Disorder

Disruptive behavior disorders such as ODD are notoriously difficult to treat. While available treatment options may include individual ther-

apy, anger management, strict behavior modification programs, boot camps, family therapy, and medication, there is limited research to support the majority of these modalities. Litschge, Vaughn, and McCrea (2009) examined 26 meta-analyses that reviewed over 2000 studies on the treatment of disruptive behavior disorders. These researchers found that among the wide variety of interventions often used, behavioral and cognitive behavioral interventions were moderately effective treating symptoms of ODD, as were family-based and multimodal therapies. In general, they summarized their findings by noting that programs that aggregate groups of youth were less effective; parent-centered programs were more effective than child-focused programs; social skills training was appropriate and helpful for many youth; and many commonly supported treatments (e.g., boot camps) are not effective. In general, research is consistent with these findings, and the general consensus is that child-focused treatments are limited in their effectiveness, and instead intervention should focus on the parent and family unit and include a variety of components such as parent management training, skill building, and multisystemic therapy (Burke et al., 2002; Capaldi & Eddy, 2015; Garland, Hawley, Brookman-Frazee, & Hurlburt, 2009; Sprague & Thyer, 2003).

Family-Based Interventions

Family-based interventions have the most empirical support in successfully treating ODD. These interventions typically involve both the parent and child throughout the course of treatment, as well as other guardians or significant family members. This type of therapy can be done in an informal manner or it may follow a more formal, structured, treatment protocol. Given that the efficacy of informal methods is difficult to follow and demonstrate in research, the following focuses on formal, evidence-based family interventions. Family-based interventions that have been empirically supported in the treatment of ODD include various types of parent management training and multisystem family therapy.

Parent Management Training Parent management training is considered a well-established evidence-based intervention for the treatment of ODD (Eyberg, Nelson, & Boggs, 2008), meaning that several research studies have found this method to be more beneficial than others in treating ODD. This treatment method focuses on teaching the parent communication, discipline, and behavior strategies that may be more effective in dealing with kids with ODD (Kazdin, 2005). The primary goal is to improve parenting skills in order to improve the parent-child relationship and subsequently reduce the child's oppositional and disruptive behavior (Feldman & Kazdin, 1995). For example, parents may be taught strategies to promote positive social behavior by utilizing techniques such as positive reinforcement like providing praise for desired behavior rather than strict punishments for unwanted behaviors (Kazdin, 2005). There are several specific types of manual-based parent training management programs (e.g., The Incredible Years, Triple P, Coping Powers, Dina Dinosaur), with a substantial body of research supporting the efficacy of these programs in a number of different settings. Research has suggested that similar to other therapy modalities, parent "readiness" to begin treatment can impact effectiveness (Andrade, Browne, & Naber, 2015), but nevertheless parent management training programs have consistently been supported in research. For example, Menting, Orobio de Castro, and Mathys (2013) conducted a meta-analysis of a specific parent training program (i.e., The Incredible Years) in which they reviewed 50 studies that used this technique. Results found that parent management training was an effective intervention in reducing disruptive behavior and defiance in all of the studies reviewed, with the effect size being largest for studies whose samples demonstrated more severe disruptive behaviors. These researchers subsequently described The Incredible Years as a well-established parent management training in treating diverse youth across settings, which is consistent with other literature.

A criticism of many clinical research studies is that, while the results are positive in research

trials, the interventions are not as effective in applied clinical or "real world" settings. While some have argued that the magnitude of effectiveness in "real world" settings may not be as great for parent management training (e.g., Michelson, Davenport, Dretzke, Barlow, & Day, 2013), a strength of this well-established method is that most research has found it to be useful even in applied clinical settings. For example, Costin and Chambers (2007) examined the effectiveness of parent management training for school-age children receiving services in a community mental health setting. These researchers followed 97 children with ODD whose parents completed at least eight parent management training sessions. Results found both clinical and statistical significance in the reduction of symptoms, with reports of reduced disruptive behaviors being provided both at the end of the parent training sessions and during a follow-up period 5 months later.

Parent-Child Interaction Training (PCIT) There is also evidence to support the effectiveness of PCIT in treating symptoms related to ODD (Burke et al., 2002; Lyon & Budd, 2010), though this treatment model is geared toward younger children with disruptive behaviors. PCIT is a family-based behavior therapy that utilizes parent management training techniques and is based on the idea of improving the parent-child relationship (Brinkmeyer & Eyberg, 2003; Eyberg, 1988). During the first phase, the parent interacts with the child in a nondirective manner while playing with the child. The purpose of this phase is to improve the child-parent relationship. In the second phase, the parent and child engage in parent-directive interaction. During this phase the parent is working on improving parenting skills such as setting clear expectation or instructions, providing praise for positive behavior, and utilizing consistent consequences such as time-out (Eyberg, 1988). Thus far, there is promising research in regard to the efficacy of PCIT in reducing behaviors related to ODD. Cooley and colleagues (2014) conducted a meta-analysis of 11 studies that utilized PCIT and found that this intervention both successfully decreased disruptive behaviors in young children from diverse populations and decreased parental stress (Cooley, Veldorale-Griffin, Petren, & Mullis, 2014). While research is promising, there are several limitations across studies. Studies have been conducted primarily in clinical settings, with limited success in replicating the findings in community-based settings. In addition, this treatment modality has been found to have high dropout rates and experience difficulties with low participant engagement (Lyon & Budd, 2010).

Multisystemic Therapy Although there is not as extensive of a body of literature to support its effectiveness, another empirically supported intervention for youth with disruptive behavior disorders is multisystemic therapy (Henggeler, Schoenwald, Borduin, Rowland, & Cunningham, 2009). This is a family-oriented therapy that is designed to be implemented in the natural environment. It is a home-based service that lasts approximately 4 months and is intended to target factors in the adolescent's social network that may be contributing to the disruptive behavior. There are several goals of multisystemic therapy, including improving parent discipline strategies, improving family communication, decreasing the youth's association with delinquent peers, improving school performance, and improving engagement in prosocial activities and support systems.

Multisystemic therapy has been found effective in treating disruptive behavior disorders such as CD and ODD. While many studies use legal involvement and delinquency as benchmarks of effectiveness (given the prevalence of ODD and CD among juvenile delinquents), studies have found evidence of both good short-term and long-term effectiveness (Butler, Baruch, Hickey, & Fonagy, 2011; Timmons-Mitchell, Bender, Kishna, & Mitchell, 2006; Welsh et al., 2012). A randomized, controlled clinical study compared multisystemic therapy in a group of 256 group of adolescents receiving standard treatment and found that it was more effective than standard treatment in reducing behaviors of ODD (Asscher, Dekovic, Manders, van der Laan, & Prins, 2013). While this and other studies have

demonstrated multisystemic therapy to be an effective treatment for ODD, it is notable that there is some debate regarding the methodology or ability to replicate findings of studies as well as the fact that there are studies demonstrating it is *not* effective in nondelinquent samples (Leschied & Cunningham, 2002; Littell, Popa, & Forsythe, 2005; Sundell et al., 2008).

Child-Focused Intervention

While individual therapy alone has limited support for effectively treating youth with ODD, behavioral and cognitive behavioral therapy may be used with these children in an attempt to decrease negative and hostile thinking, while also working to improve social and problem solving skills. Although cognitive behavioral therapy has limited evidence to support its effectiveness when used in isolation, some have proposed that this child-focused therapy can be beneficial when working with older youth, particularly if parent training strategies are also implemented (Eyberg et al., 2008). Skills that are often targeted in individual therapy include anger management and flexible thinking in hopes to decrease the child's emotional overreactions and ongoing defiance. Problem solving skill building is also often encouraged during individual therapy, with the goal being to improve reasoning and problem solving skills with parents and authority figures rather that arguing (Kazdin, 2003). While there are limited research studies available to support the effectiveness of cognitive behavior or other individual therapies in reducing symptoms of ODD as well or better than standard treatment or well-established treatments, Garland and colleagues (2008) did provide a review of common elements of individual treatments that can improve effectiveness for children with disruptive and defiant behaviors. These included factors such as therapeutic components (e.g., positive reinforcement strategies, teaching problem solving skills), therapeutic techniques (e.g., using role playing and assignment homework), as well as extraneous factors such as the therapist-child relationship and duration of treatment. Ultimately, these researchers concluded that individual treatments can be integral if they include components key to success (Garland, Hawley, Brookman-Frazee, & Hurlburt, 2008).

There are no medications specifically targeted toward treating symptoms of ODD. In addition, there are few empirical studies available that address the efficacy of using medications, and guidelines from the American Academy of Child and Adolescent Psychiatry indicate that medication alone is not considered a sufficient treatment for ODD. That being said, given the high comorbidity between ODD and ADHD, some professionals have argued that symptoms of ODD often decrease when psychopharmacological treatment is used for ADHD (Turgay, 2009). It has also been suggested that mood stabilizers may be beneficial in treating aggression and related symptoms when ODD is comorbid with CD (Malone, Delaney, Luebbert, Carter, & Campbell, 2000); Turgay, 2009).

Conclusion

Oppositional defiant disorder is a disruptive behavior disorder used to classify children and adolescents who demonstrate significant levels of defiance and noncompliance. Irritability, defiance, and argumentative behavior is common in all children to some degree, so ODD is a diagnosis appropriate only when these behaviors occur to such a degree that they interfere with academic success, peer relationships, familial relationships, or other major areas of life. While having a diagnosis ODD does not imply a child or adolescent lacks empathy, is likely to develop antisocial personality disorder or have significant interpersonal difficulties throughout life, ODD is associated with several risk factors and serious implications (e.g., school failure, involvement with the juvenile justice system). In addition, ODD and other disruptive behavior disorders can cause significant distress and be related to dysfunction in the family environment.

While a variety of intervention and treatment modalities are available, few have strong evidence to support their efficacy and effectiveness

in everyday settings, and ODD remains a disruptive behavior disorder that is relatively difficult to treat. At the time of this publication, parent management training techniques are the only well-established treatment for ODD. There are a variety of other promising interventions, but further research and clinical trials are needed to determine the effectiveness. Treatment difficulty does not imply that ODD cannot be treated; rather, difficulties developing effective treatment programs are likely due to several factors. First, there is some controversy regarding the actual diagnostic process of ODD and being able to reliably and validly diagnose it in children and adolescents. Diagnostic criteria have been refined and manipulated with each revision of the DSM, and this is likely to continue as we better understand the disorder and how it presents in different individuals. Second, in addition to diagnostic considerations, treatment difficulties are likely related to the fact that there is no one specific etiology or causal factor related to the development of ODD. Rather, at the present time it appears that there are a variety of biological and environmental factors that contribute to its development. This makes for more complex treatment programming as the various contributing factors for each youth need to be identified, and it suggests that progress in intervention and treatment programs may be slow as various settings and until researchers can identify biological markers or major environmental factors across youth with ODD. Finally, treating children and adolescents with disruptive behavior disorders is likely to always be more difficult because these youth spend considerable time in different settings (i.e., home and school), with different caregivers and authority figures. Thus far there is less benefit observed in child-focused treatments, so while techniques such as parent management training may be highly successful, results may still be limited as the skills the parent learns cannot necessarily be utilized during the 8 h each day the child is at school.

Significant progress in understanding and treating ODD has been made since it was first included in the DSM-II; however, our understanding of this disorder and the ability to quickly and effectively treat symptoms are still somewhat archaic in nature compared even to other disruptive behavior disorders such as ADHD. As the field of cognitive neuroscience grows, we can hope that our understanding of biological factors of ODD will grow, along with our understanding of specific environmental contributions to the disorder. In addition, with the growing emphasis on evidence-based practice, it can be expected that treatment for ODD will continue to be refined and more well-established intervention methods will become available.

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Conduct Disorder

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Introduction

Antisocial behavior has a profound psychological and social impact on both the individual committing such acts and society at large. Thus, understanding and preventing severe antisocial behavior among children and adults is an important challenge faced by modern societies. Conduct disorder (CD) is a common childhood psychiatric disorder that has an increased incidence in adolescence and is characterized by "a repetitive and persistent pattern of behavior by a child or teenager in which the basic rights of others or major age-appropriate societal norms or rules are violated" (p. 469, American Psychiatric Association [APA], 2013). The primary diagnostic features of CD include aggression to people or animals, destruction of property, deceitfulness or theft, and serious violations of rules (APA, 2013). CD is known to have a significant impact on individuals, their families, and society, and is a risk factor for school dropout, delinquency, and teenage pregnancy (Hill, 2003). Severe behavioral problems in children can lead to more serious offending patterns as children develop, which place a huge financial burden on society (Romeo, Knapp, & Scott, 2006; Scott, Knapp, Henderson, & Maughan, 2001). Indeed, by the time they are 28 years old, individuals diagnosed with CD before age 10 cost society ten times as much as individuals who did not have conduct problems (CP) as children (Scott et al., 2001). In the USA, annual healthcare costs for children with CD are $10,000 more than healthcare costs for healthy children (Merikangas, Nakamura, & Kessler, 2009), whereas estimates in the UK report the overall care of a child referred for persistent antisocial behavior costs at nearly £6000 per annum (Romeo et al., 2006). Importantly, efforts to imprison and monitor parolees are perceived as more expensive and time-consuming than prevention efforts used to identify early risk factors and tailor intervention for children with CD (Hill, 2003).

The prevalence, as well as the cost, of CD in our society has prompted scientists and clinicians to investigate and identify its characteristics, predictors, and causes in the hope of developing effective interventions and preventions. This chapter will discuss the diagnosis of CD in terms of the most recent edition of the *Diagnostic and Statistical Manual* (DSM-5) (APA, 2013). First, a brief historical overview of the diagnosis will be provided, followed by a description of the current diagnostic criteria as well as prevalence in community, clinical, and forensic settings. The second section will focus on the clinical presentation with an emphasis on the age of onset, level of callous-unemotional traits, sex differences, and comorbidities. The third section will critically discuss environmental risk factors for CD as well as its neurobiological correlates

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identified through genetics, epigenetics, autonomic nervous system responsivity, levels of neurotransmitters, neuropsychological performance, and structural and functional neuroimaging. The chapter will conclude by reviewing evidence-based treatment options, prognosis, and outcome measures among subgroups of children with CD.

Overview and History

Prior to its recognition as such, CD was first identified in the DSM-II as "socialized aggressive reaction of childhood" and characterized by overt or covert hostile disobedience, quarrelsomeness, physical and verbal aggression, vengefulness, and destructiveness (APA, 1968). Temper tantrums, solitary stealing, lying, and hostility toward other children were also identified (APA, 1968). The DSM-III officially changed "socialized aggressive reaction of childhood" to CD and introduced four subtypes: socialized, socialized/psychopathic, neurotic, and inadequate/immature (APA, 1980); (Adam, Kashani, & Schulte, 1991). These subtypes were abandoned in the DSM-IV, which introduced the age-based distinction between childhood-onset and adolescent-onset CD (APA, 1994); (Moffitt, 1993; Moffitt, Caspi, Dickson, Silva, & Stanton, 1996). The most recent edition, DSM-5 (APA, 2013), kept the age-based distinction, but introduced the "with limited prosocial emotions" specifier to further distinguish youths with CD as those displaying high (CD/HCU) versus low (CD/LCU) callous-unemotional (CU) traits. This subtyping approach is based on the extensive body of research indicating that HCU traits identify a particularly problematic subgroup of youths with CD with more severe antisocial and aggressive behavior than other youths with CD (Frick, Ray, Thornton, & Kahn, 2014b). CU traits reflect a lack of empathy and guilt combined with a shallow affect and the callous use of others for one's own gain (Frick & White, 2008). The inclusion of CU traits as a specifier in the DSM-5 reflects a departure from subtype approaches adopted in previous versions of the DSM that were mostly based on observable behavior rather than personality traits (Frick & Marsee, 2006; Frick & White, 2008).The inclusion of the "with limited prosocial emotions" specifier may also help researchers and clinicians to better diagnose CD in this heterogeneous population and aid effective treatment and prognosis (Hawes, 2014). Differences in the presentation of CD based on age of onset and severity of CU traits are discussed further below.

DSM-5 Diagnostic Criteria and Presentation

The DSM-5 describes CD as a childhood psychiatric problem that has an increased incidence in adolescence (APA, 2013). The primary diagnostic features of CD are severe externalizing behaviors including aggression to people or animals, destruction of property, deceitfulness or theft, and serious violations of rules. To receive a diagnosis of CD, a child must present with 3 out of 15 possible symptoms within the past 12 months and with 1 symptom present within the past 6 months (APA, 2013; see Table 26.1). Common symptoms include lying multiple times per week with the intention to manipulate or deceive others, truancy on more than two occasions, staying out late or overnight without permission on multiple occasions, and engaging in fighting or intimidating peers or adults. Children with CD often engage in impulsive and risky activities and regularly have temper outbursts and/or defiant attitudes toward authority figures (Loeber, Burke, Lahey, Winters, & Zera, 2000). These children also struggle with delayed gratification relative to typically developing (TD) children, and they tend to choose immediate rewards over larger, delayed rewards (White et al., 2014). CD can be classified as mild, moderate, or severe based on the type and severity of symptoms and the level of harm to others (APA, 2013). A diagnosis is made when the disturbance in behavior causes clinically significant impairment in social, familial, academic,

Table 26.1 DSM-5 conduct disorder diagnostic criteria

Symptoms	Criteria
Aggression to people and animals	1. Often bullies, threatens, or intimidates others 2. Initiates physical fights 3. Has used a weapon to cause serious harm to others 4. Has been physically cruel to people 5. Has been physically cruel to animals 6. Has stolen while confronting a victim 7. Has forced someone into sexual activity
Destruction of property	8. Has engaged in fire setting with intention to damage 9. Has deliberately destroyed other property (other than fire setting)
Deceitfulness or theft	10. Has broken into a house, car, building 11. Lies to obtain goods or avoid obligations 12. Has stolen items of nontrivial value without confronting a victim (shoplifting, forgery)
Serious violation of rules	13. Stays out at night despite parental prohibitions, beginning before 13 years 14. Has ran away from home overnight at least twice or once when staying away for a lengthy period of time 15. Often truants from school, beginning before 13 years

or occupational functioning. The fictionalized vignette below gives an example of the types and combinations of symptoms seen in a youth with moderate CD.

Troy exhibits 5 of the 15 symptoms of CD, beginning at the age of 8 where he stole items of nontrivial value without confronting a victim (#12) and subsequently lied about it (#11). At middle school, he continued to steal without confrontation, bullied other males (#1), skipped school (#15), and set fire to public property (#8). Once he turned 18, he continued to behave antisocially, culminating in a prison sentence for assault. Troy received a childhood-onset CD diagnosis, as at least one of the symptoms was present before the age of 10.

Case Study: Troy

Troy lives with his mom, dad, and five younger siblings in a four-bedroom house in an urban city. His mother smoked throughout pregnancy and was 21 years old when Troy was born. Troy's parents had a turbulent relationship; his father left the family and then returned on several occasions. Troy's teachers commented on his difficult temperament and issues on sharing and cooperating with other children during preschool. Troy's mother started noticing that he would steal food and occasionally money from her purse when he was 8 years old. He would never admit to it when challenged and was screamed at by his mom whenever he was caught. He struggled in elementary school and had difficulties making friends. When he moved to middle school, he made new, older friends who introduced him to smoking. His schoolwork rapidly went downhill as he began to truant and argue with teachers. He stole cigarettes and money from his parents in order to impress his friends and bullied younger children into stealing money for him. Troy had many girlfriends, with whom he formed shallow, short-lived relationships, and was sexually active from 14 years old. He became known to the criminal justice system when he set fire to a seat on the bus with a couple of friends at 15 years old. His parents were emotionally distant from Troy, and he played up to get their attention. His teacher suggested he should be assessed for ADHD, but he missed the appointment with the psychiatrist, and at age 16 he dropped out of school. As he had nothing to do, he hung around more with his older friends who encouraged him to take drugs. At 17, his parents kicked him out of the house and he had no stable home, working sporadically and making money from illegitimate means. He fathered a son at 18, who was born while Troy was serving time in prison for assault. Troy was diagnosed with antisocial personality disorder at 25 years old.

Prevalence

CD has a prevalence of approximately 9.5% in the USA (Nock, Kazdin, Hiripi, & Kessler, 2006) and 5.8% in the UK (Green, McGinnity, Meltzer, Ford, & Goodman, 2005). Rates of CD are double among males in comparison to females (Maughan, Rowe, Messer, Goodman, & Meltzer, 2004). Discrepancies across countries may reflect a true difference or may reflect difference in methodologies used to assess prevalence (De Brito & Hodgins, 2009). CD and associated disruptive behavior disorders (DBD) are the most common reason for child and adolescent referrals to mental health clinics. Nearly 50% of child health referrals are for DBD and 30% of referrals to pediatricians are for DBD (NICE, 2013). Furthermore, mental illness has been recognized in over half (53%) of children in juvenile detention centers with 40% of those children meeting diagnostic criteria for DBD including oppositional defiant disorder (ODD) and CD (Shelton, 2001).

Clinical Presentation

Childhood Onset vs. Adolescent Onset

One of the major criticisms of the diagnosis of CD is that it primarily indexes overt antisocial behavior, which results in the identification of a highly heterogeneous group of children with different temperamental and behavioral characteristics (Blair, Peschardt, Budhani, Mitchell, & Pine, 2006); (Frick & Marsee, 2006). It has been proposed that these differences reflect distinct developmental pathways to CD (Frick & White, 2008). In the DSM-IV, this heterogeneity among CD was acknowledged, with the age-based distinction between childhood-onset CD (i.e., presence of at least one symptom before age 10, such as Troy) and adolescent-onset CD (i.e., absence of any symptom before age 10). The age-based distinction has been well supported for both females and males, particularly in terms of its predictive validity and its ability to identify the characteristic problems of the two subtypes (Moffitt et al., 2008). Age of onset of CD predicts chronic delinquency (Moffitt, 2006), and it is one of the best predictors of antisocial behavior in adulthood (Loeber, 1991). The younger the age of onset of CD is, the greater the number of aggressive behaviors and the higher the level of functional impairment (Lahey et al., 1999). Additionally, a longitudinal study has shown children with the greatest number of antisocial behaviors before the age of 10 are more likely to have difficulties functioning in society by their mid-twenties (Fergusson, John Horwood, & Ridder, 2005).

More than two decades of research have identified important differences between youths with childhood- and adolescent-onset CD. Those with childhood-onset CD have a higher genetic liability for antisocial behavior, more frequently present with family dysfunction, antisocial parents, neurocognitive impairments such as lower IQs, and higher levels of comorbidities such as hyperactivity and inattention disorders compared to youths with adolescent-onset CD (Moffitt et al., 2008). These impairments are reflected in higher levels of school difficulties and peer relation problems. The adolescent-onset subtype, by contrast, tends to exhibit fewer difficulties in these areas (Moffitt et al., 2008). Research suggests that risk-taking and rebellious behavior increase during teenage years within the general population and then decline in adulthood (Steinberg, 2004). However, youths with adolescent-onset CD exhibit a higher frequency and severity of CD symptoms compared to TD adolescents. While childhood-onset CD is thought to have a neurocognitive basis, it has been posited that adolescent-onset CD is primarily caused by association with other delinquent youths (Moffitt, 1993). This distinction has, however, recently been criticized by Fairchild et al. (2013) who suggested that differences between the two subtypes are quantitative rather than qualitative. Specifically, Fairchild and colleagues concluded that neurocognitive deficits are present in both subtypes and that environmental factors act as moderators between a child's predisposition and age of onset (Fairchild et al., 2013).

High vs. Low Callous-Unemotional Traits

An extensive body of research has highlighted the theoretical and clinical importance of distinguishing between youths with CD/HCU and those with CD/LCU (Frick et al., 2014c). CU traits are a central feature of the adult syndrome of psychopathy (Cleckley, 1982), and youths with CD/HCU are thought to be at an increased risk of developing psychopathy in adulthood (Blair, Leibenluft, & Pine, 2014; Frick, Ray, Thornton, & Kahn, 2014a). CU traits in late childhood and adolescence are positively correlated with psychopathy scores in adulthood, even after controlling for childhood CD and other childhood risk factors (Burke, Loeber, & Birmaher, 2002); (Lynam, Caspi, Moffitt, Loeber, & Stouthamer-Loeber, 2007). There is overwhelming evidence that HCU traits identify a particularly problematic subgroup of youths with CD, who show a more severe, stable, and aggressive pattern of antisocial behavior than other youths with CD (Frick et al., 2014b), leading to the inclusion of the "with limited prosocial emotions" CD specifier within DSM-5 (APA, 2013). To qualify for this specifier, the individual has to meet full criteria for CD as well as persistently present with two of the following four characteristics in multiple relationships and settings over a minimum 12-month period: lack of remorse or guilt, callous-lack of empathy, unconcern about performance at school or work, and shallow or deficient affect. A recent study examining the prevalence of this CU traits specifier among youths reported that in a community sample, 10–32% of those with CD met the CU specifier threshold, while these figures increased to 21–50% of those with CD from a clinical sample (Kahn, Frick, Youngstrom, Findling, & Youngstrom, 2012). Interestingly, across both community and clinical samples, between 2% and 32% of youths without CD also met diagnostic threshold for the CU specifier, but those with CD and CU traits showed consistently higher rates of aggression and cruelty (Kahn et al., 2012) (for a review, see Herpers, Rommelse, Bons, Buitelaar, & Scheepers, 2012).

Youths with CD/HCU and CD/LCU show distinct temperamental and affective profiles. Those with CD/HCU traits are insensitive to punishment but are more receptive to reward, consistent with a preference for novel and dangerous activities and impulsive behavior (Blair, 2013; Frick & Marsee, 2006). In addition, they underestimate the likelihood of being punished for their actions relative to youths with other behavioral problems (Pardini, Lochman, & Frick, 2003). In contrast to youths with CD/LCU traits, those with CD/HCU traits show a weaker response to distress cues in others (Blair, 1999; Blair, Colledge, Murray, & Mitchell, 2001; Blair & Frith, 2000; Marsh et al., 2011) and impairment in emotion recognition (Fairchild, Van Goozen, Calder, Stollery, & Goodyer, 2009; Sharp, Vanwoerden, Van Baardewijk, Tackett, & Stegge, 2014). There is also evidence that HCU traits are associated with lower levels of anxiety and fear, especially when controlling for impulsivity and CP (Frick, Lilienfeld, Ellis, Loney, & Silverthorn, 1999; Lynam et al., 2005; Pardini & Frick, 2013). By contrast, youths with CD/LCU traits do not show comparable punishment or reward sensitivities, but have problems regulating their emotions, displaying increased levels of anger and impulsivity (Frick & Marsee, 2006; Frick et al., 2014b). They are also more attuned to negative stimuli (Viding & McCrory, 2012b) and to the distress of others (Jones, Happe, Gilbert, Burnett, & Viding, 2010).

Given those distinct profiles, it is unsurprising that youths with CD/HCU traits and those with CD/LCU present with distinct patterns of antisocial and aggressive behavior. The association between an early (i.e., school age and early adolescence) stable presentation of antisocial behavior and CU traits (Dandreaux & Frick, 2009; Frick & Viding, 2009; Rowe, Costello, Angold, Copeland, & Maughan, 2010) has been linked to higher levels of aggression (Cornell et al., 1996; Frick, Cornell, Barry, Bodin, & Dane, 2003) over and above correlations with rule-breaking behaviors (e.g., Poythress, Dembo, Wareham, & Greenbaum, 2006). In a sample of high-risk males, HCU traits predicted an increased likelihood of violent and aggressive offending as

an adult, even when controlling for age of onset (Loeber et al., 2005). Furthermore, compared to antisocial youths with LCU traits, those with HCU traits displayed a more varied and severe pattern of aggressive behaviors, including instrumental and premeditated aggression, akin to those observed in adult psychopathy at a younger age (Frick, Cornell, Bodin, et al., 2003; Frick & Marsee, 2006; Frick et al., 2014b). By contrast, youths with LCU traits have typically been found to be less aggressive, displaying predominantly threat-based reactive aggression (Viding & McCrory, 2012b).

In sum, these findings support the view that the presence of CU traits designates a distinct group of children and adolescents who show a particularly severe, aggressive, and stable pattern of antisocial behavior (Frick, Ray, Thornton, & Kahn, 2014a; Frick et al., 2014c; Frick & White, 2008). Importantly, the positive association between CU traits and severity of antisocial and aggressive behavior has been observed for both males (Kruh, Frick, & Clements, 2005) and females (Marsee & Frick, 2007) and in children as young as 2 (Waller et al., 2012) and 3 years old (Hyde et al., 2013; Kimonis et al., 2006). It must be noted, however, that results are often correlational, and so any observed associations between CU traits and rates of aggression and cruelty cannot be considered causal. It is possible, for instance, that while high levels of CU traits can lead to increased aggression, a history of early adversity could also make someone more callous and unemotional (see Kumsta, Sonuga-Barke, & Rutter, 2012).

Sex Differences

There are well-documented sex differences in the prevalence of CD, which is more frequently diagnosed in males (Maughan et al., 2004), with a sex ratio of about 2:5 females to males (Moffitt & Caspi, 2001). The basis of these sex differences in the prevalence of CD remains unclear. Originally thought of as cultural bias (Robins, 1991), there has been much debate as to whether the diagnostic criteria for CD are tailored to fit predominantly overt antisocial male behaviors (e.g., aggression, theft, vandalism, rule breaking) rather than more covert antisocial female behaviors (e.g., deceit, manipulation, bullying, lying, staying out late) (Leadbeater, Kuperminc, Blatt, & Hertzog, 1999; Moffitt et al., 2008). For instance, among a community sample of young females with CD, the most common symptom reported by parents was lying to deceive others (Hipwell et al., 2002). As most female antisocial behavior occurs in adolescence (Fontaine, Carbonneau, Vitaro, Barker, & Tremblay, 2009; Moffitt & Caspi, 2001), some researchers suggest that age of onset in females with CD is restricted to adolescence (Silverthorn & Frick, 1999). However, a more recent longitudinal study has shown that while females are more prone to develop adolescent-onset CD, they do in fact show both childhood- and adolescent-onset CD (Odgers et al., 2008).

CD in females has received increased attention recently following evidence documenting increased rates of CD diagnoses in the USA and UK and number of arrests for crimes perpetrated by females in the past 10 years (Collishaw, Maughan, Goodman, & Pickles, 2004; Hawkins, Graham, Williams, & Zahn, 2009). Addressing the increased prevalence of CD in females is of paramount importance given its impact on social adjustment and implications for development of unhealthy interpersonal relationships with partners and offspring (Keenan, Loeber, & Green, 1999). For example, females with CD tend to obtain lower levels of education and/or become a single parent at a younger age than their TD peers (Hill, 2003). Additionally, females with CD are more likely to have antisocial partners and raise children with behavioral problems (Keenan et al., 1999). They are also more likely to partake in risky activities, become targets for grooming and prostitution, and are more susceptible to drug abuse and contracting sexually transmitted diseases (Bardone et al., 1998; Pedersen & Mastekaasa, 2011).

Females with CD show unique patterns of aggression, showing more relational or social aggression (e.g., gossiping, spreading rumors, and exclusion from social groups), than TD males (Crick & Grotpeter, 1995; Hipwell et al.,

2002). Females also tend to display direct or overt aggressive behavior when they are younger and more indirect or covert aggressive behavior during early adolescence; this switch in the type of aggression typically occurs earlier in females than in males (Wolke, Woods, Bloomfield, & Karstadt, 2001). Females with CP also present with lower levels of aggression but cause less damage and physical harm to other people than males with CP (Lahey et al., 1998). In forensic settings, females held in juvenile detention centers tend to be more relationally aggressive and less physically aggressive than males (Marsee & Frick, 2007). Among these females, those with high levels of CU traits are more likely to exhibit aggressive behavior in social situations than those with LCU traits, who tend to exhibit reactive aggression socially (Marsee & Frick, 2007).

Comorbidities

It is a goal of this chapter to highlight that youths with CD are a very heterogeneous population. This is largely due to the fact that CD rarely occurs singly (Turgay, 2005); youths with CD often also present with other externalizing disorders, such as attention-deficit hyperactivity disorder (ADHD), oppositional defiant disorder (ODD), and substance use disorder, as well as internalizing disorders such as mood and anxiety disorders (Maughan et al., 2004). There is now good evidence that CD begins before most of its comorbid disorders (Nock et al., 2006). Females with CD can often be misdiagnosed, due to the high rate of comorbidities between CD and other disorders (Euler et al., 2014). While females with CD are more prone than males with CD to internalizing disorders such as depression, anxiety, and eating disorders, males with CD are more likely to present externalizing disorders including ADHD, ODD, and substance abuse (Leadbeater et al., 1999). There is also evidence that covariation between CD and substance misuse reflects a common latent externalizing factor that is heritable (Krueger, Markon, Patrick, Benning, & Kramer, 2007) and that common genes also influence the co-occurrence between externalizing and internalizing problems (Kuja-Halkola, Lichtenstein, D'Onofrio, & Larsson, 2015). These aspects further complicate diagnosis and treatment, while also making it difficult for researchers and clinicians to disentangle the biological variations exclusively related to CD.

ADHD

ADHD is a neurodevelopmental disorder characterized by continuous lack of attention or concern, impulsive behavior and decision-making, and inability to plan and organize and is first noticeable in early childhood (APA, 2013). It is also one of the most likely to co-occur alongside CD (Loeber & Keenan, 1994); between 30% and 50% of children with ADHD also have CD according to both epidemiological and clinical samples (for review see Deault, 2010), with a common genetic liability believed to account for ADHD and childhood-onset CD (Silberg, Moore, & Rutter, 2015). A lack of positive maternal involvement, which is often seen in parents of children with CD (Frick et al., 1992), may exacerbate both ADHD and CD (Pfiffner, McBurnett, Rathouz, & Judice, 2005). Children with both CD and ADHD have been shown to be more aggressive and have a poorer adult prognosis than those with CD alone (Lahey, Loeber, Burke, & Applegate, 2005). Youths with CD and ADHD, compared to those with CD alone, are also characterized by an earlier age of onset for behavioral problems, lower verbal and social-cognitive abilities, and increased problems with peers (Lahey et al., 2005; Lynam, 1996; Waschbusch, 2002). In line with evidence suggesting that these disorders are heterogeneous in terms of neurocognitive impairments (Rubia, 2011), various studies have shown that children with ADHD alone are not at higher risk for demonstrating antisocial behavior in adulthood, while those with comorbid CD and ADHD are more likely to present with persistent antisocial activities beyond childhood (Mannuzza, Klein, & Moulton, 2008; Mordre, Groholt, Kjelsberg, Sandstad, & Myhre, 2011).

Oppositional Defiant Disorder

ODD is marked by an angry, irritable mood, argumentative behavior, avoidance of prosocial behavior, and vindictiveness. It is strongly linked to emotion dysregulation and is less severe than CD (APA, 2013). A large, cross-sectional study has found that a significant proportion of males with CD have a prior diagnosis of ODD, unlike most females with CD who do not (Lahey et al., 2000).

ODD was originally thought to be a precursor for CD (Biederman et al., 1996), which prevented simultaneous presentation of both ODD and CD. However, recent research has suggested that these disorders are discrete and should be considered as such (Althoff, Kuny-Slock, Verhulst, Hudziak, & van der Ende, 2014; Burke, Waldman, & Lahey, 2010; Pardini, Frick, & Moffitt, 2010); hence the DSM-5 now lists them as separate disorders and allows for comorbidity (APA, 2013). Rowe et al. (2002) compared the overlap between ODD and CD and found that most males with a diagnosis of CD showed oppositional features, with only 5% showing no characteristics and 25% meeting full criteria for ODD. A stronger overlap was observed in females, where only 2% of females with CD showed no ODD symptoms and 54% met full ODD criteria. It has been suggested that environmental factors such as parenting and low SES contribute to externalizing behaviors more often in cases of comorbid CD/ODD but not when CD is present on its own (Rowe et al., 2010).

However, it is unclear whether ODD is a discernible disorder or whether it is a nonspecific marker for maladjustment (Frick & Nigg, 2012). For example, many TD children show oppositional behaviors during healthy development (Wakschlag et al., 2007), and ODD is often comorbid with various other disorders (Rowe et al., 2002). Therefore, severe oppositional behaviors may be symptomatic of other disorders, rather than a disorder itself. However, Frick and Nigg (2012) argue that ODD retains important diagnostic utility, as it predicts for problems in adjustment, emotional disorders (Loeber, Burke, & Pardini, 2009), and functional impairment in adulthood (Burke et al., 2010).

Substance Use Disorders

Children and adolescents with CD are more likely to be exposed to alcohol and illicit drug use at a young age than TD children (Cadoret, Yates, Troughton, Woodworth, & Stewart, 1995). Subsequently, those with CD are more likely to use substances (Sung, Erkanli, Angold, & Costello, 2004) and are at a higher risk of developing substance use disorders compared to both TD youths and those with pure ADHD (Costello, Mustillo, Erkanli, Keeler, & Angold, 2003; Disney, Elkins, McGue, & Iacono, 1999). Copur, Turkcan, and Erdogmus (2005) found that 80 out of 230 juvenile prisoners abused substances, and approximately 45% of adolescents who abused substances also suffered from CD (Copur et al., 2005; Langenbach et al., 2010). Some health outcomes associated with CD such as sexually transmitted diseases are also highly related to substance use disorders (Courtney & Polich, 2009), with twin studies suggesting that genetic influences associated with CD often overlap with genetic vulnerability for alcohol and other substance dependence (Kendler, Prescott, Myers, & Neale, 2003; Slutske et al., 1998).

Internalizing Disorders

While some had suggested that the presence of internalizing disorders, such as anxiety disorders, reduced the severity of CD (e.g., Walker et al., 1991), recent research indicates high comorbidity levels between CD and internalizing disorders. Two decades ago, a review reported that between 22% and 33% of youths in community samples presented with comorbid CD anxiety disorders (Russo & Beidel, 1994), but more recent large cross-sectional and longitudinal studies have found that between 7% and 13% of those with CD have an anxiety disorder, while between 12% and 17% also have a depressive disorder (Maughan et al., 2004; Rowe, Maughan, Costello, & Angold, 2005). Another recent study, which recruited a clinical and a community sample with undiagnosed conduct problems, estimated that 78% of children and adolescents from the clinical sample and 35% of those from the community sample have a comorbid internalizing disorder (Polier, Vloet, Herpertz-Dahlmann,

Laurens, & Hodgins, 2012). One possible reason for the discrepancy between those estimates could be due to the fact that some studies used parent-rated questionnaires (Polier et al., 2012), while others used interviews and ratings by clinicians (Rowe et al., 2005).

Environmental Risk Factors

A large body of evidence from cross-sectional and longitudinal epidemiological studies has accumulated regarding the environmental risk factors for CD (Latimer et al., 2012), which include, among others, maternal smoking (Slotkin, 2013) and drug use during pregnancy (Amaro, Zuckerman, & Cabral, 1989; Frick et al., 1992) and other perinatal-related risk factors (Moffitt, 1993), malnourishment (Raine, Mellingen, Liu, Venables, & Mednick, 2003), low SES and poverty (Odgers et al., 2012), neighborhood disadvantage (Loeber, Green, Keenan, & Lahey, 1995), maternal and paternal psychopathology (Barker, Copeland, Maughan, Jaffee, & Uher, 2012), acute and chronic experience of trauma including maltreatment and low parental warmth (for a review see McCrory, De Brito, & Viding, 2010), and association with delinquent peers (Moffitt, 2006).

Finally, migration status has been reported as a risk factor for CD in the USA (Breslau et al., 2011) but has not been studied as a risk factor in Europe. Of the risk factors listed, SES, education, and occupation of parents are among the strongest predictors of behavioral problems (Bradley & Corwyn, 2002). Interestingly, a recent study reported that by age 12, the effect of neighborhood SES on children's antisocial behavior was as large as the effect observed for sex, which is considered the most robust predictor of antisocial behavior (Odgers et al., 2012). Poor nutrition and malnourishment have also been linked to behavioral problems, with omega-3 supplementation found to reduce both externalizing and internalizing problems (Raine et al., 2003). While association with deviant peers is thought to be a robust risk factor for CD (Javdani, Sadeh, & Verona, 2011), this may be dependent on sex according to one large longitudinal study, which found that socialization with the opposite sex was associated with increased violence in females, but decreased violence in males (Haynie, Steffensmeier, & Bell, 2007). Finally, another longitudinal study found that while sexual abuse alone predicts adolescent delinquent behavior, co-occurring neglect and physical and sexual abuse are the greatest predictors of delinquent behavior (Hahm et al., 2010).

Neurobiological Factors

There is an extensive literature on the neurobiological correlates of CD and its subtypes (Blair, 2013; Fairchild et al., 2013; Matthys, Vanderschuren, & Schutter, 2013). Here, we briefly review and critically discuss the evidence pertaining to heritability and candidate genes, epigenetics, autonomic nervous system, levels of neurotransmitters, neuropsychological test performance, as well as more recent functional MRI and structural MRI studies.

Heritability and Candidate Genes

Estimates from over 100 quantitative genetic studies suggest that genes contribute around 50% of variance in CD (Moffitt, 2005). Although there is clear evidence for a genetic influence on CD, few studies have looked at isolating the precise genes responsible (Dick et al., 2011). Genes thought to be pertinent in CD include those associated with neurodevelopment (Dadds, Moul, Cauchi, Hawes, & Brennan, 2013), dopamine (Beaver et al., 2007), serotonin (Iofrida, Palumbo, & Pellegrini, 2014), and gamma-aminobutyric acid (GABA) regulation (Dick et al., 2006; see Iofrida et al. (2014) for a review). While adolescent-onset CD has been posited to be associated with peer relationships (Moffitt et al., 1996), childhood-onset CD is considered to be caused by a combination of genetic factors and environmental risk factors (Moffitt et al., 1996). However, recent research has not supported this claim, with reporting that genes account for 62% of childhood-onset CD and 65% of adolescent-onset CD variability.

With regard to CU traits, Viding, Jones, Frick, Moffitt, and Plomin (2008) replicated and

extended an earlier report on 7-year-old twins (Viding, Blair, Moffitt, & Plomin, 2005) by showing that when hyperactivity symptoms were controlled for, genetic factors accounted for 71% of the variance associated with antisocial behavior in 9-year-old twins exhibiting CP/HCU traits, but only 36% of the variance in those exhibiting CP/LCU traits. Heritability estimates with regard to sex are inconsistent across the literature; a study comparing genetic influences in a community sample of male and female monozygotic and dizygotic twins found that genetic factors may have a larger influence in females than in males when considering *nonaggressive* antisocial behavior; however, no difference was found for *aggressive* antisocial behavior (Eley, Lichtenstein, & Stevenson, 1999). More recently, Baker, Raine, Liu, and Jacobson (2008) found moderate genetic influences for males (but not females) and moderate shared environmental influences for females (but not males), with regard to aggressive behavior. However, a contrasting study reported that while genetics and environmental influences had the same amount of influence (i.e., no quantitative sex difference) on antisocial behavior in adults, genetic or shared environmental factors influenced antisocial behavior differently in males and females in childhood, suggesting qualitative differences between the sexes (Meier, Slutske, Heath, & Martin, 2011). The discrepancy in these studies could be due to, among other factors, the way antisocial behavior is operationalized; males are more likely to display overt/physical aggression, while females are more likely to present relational aggression (Crick & Grotpeter, 1995; Ostrov & Keating, 2004). Based on the results of a handful of studies, CU traits among males seem to be more heritable than among females (Viding & McCrory, 2012a), but additional studies are needed.

A number of candidate genes have been investigated with regard to CD and antisocial behavior, including those involved in dopaminergic (Guo, Roettger, & Shih, 2007) and serotonergic (Ficks & Waldman, 2014) circuits (see Iofrida et al. (2014) for comprehensive review). Using a nationally representative community sample of 2500 adolescents, Guo et al. (2007) reported a main effect on delinquency of the TaqI polymorphism in the DRD2 gene and of the 40 bp VNTR polymorphism in the DAT1 gene. Specifically, Guo and colleagues found that adolescents with the DAT1*10R/10R and DAT1*10R/9R genotypes were more likely to show antisocial behavior than those with the DAT1*9R/9R genotype. Furthermore, those with heterozygotic genes (A1/A2) for DRD2 were more likely than those with homozygotic genes (both A2/A2 and A1/A1) to show delinquent behavior. Interestingly, neither variant was associated with delinquency in females. Crucially, however, genetic variations in some genes interact with environmental factors. One example is the monoamine oxidase A (MAOA) gene, which degrades serotonin pre- and post-synaptically (Buckholtz & Meyer-Lindenberg, 2008). The MAOA gene was first thought to be associated with antisocial behavior when a mutation in this gene was found in a Dutch family with a history of severe aggression and violence spanning several generations (Brunner et al., 1993). Subsequent research has concluded that the deletion of MAOA gene produces aggressive behaviors in humans and in animals (Alia-Klein et al., 2008). There is now good evidence that variations in the MAOA gene also interact with maltreatment to influence the development of CD and Antisocial Personality Disorder. In a seminal study, maltreated children carrying the low-activity allele, which confers low levels of MAOA expression, were more likely to develop CD and ASPD than those carrying the high-activity allele, which is associated with high levels of MAOA expression (Byrd & Manuck, 2014; Caspi et al., 2002).

While, to our knowledge, no research has examined candidate genes in relation to age of onset in CD, a number of studies have investigated main effects of genetic variants on CU traits (Moul, Dobson-Stone, Brennan, Hawes, & Dadds, 2013) as well as their interactions with environmental factors (Sadeh et al., 2010; Willoughby, Mills-Koonce, Gottfredson, & Wagner, 2014). For example, Sadeh et al. (2010) explored the influence of a polymorphism in the serotonin transporter protein gene

(SLC6A4) and socioeconomic resources (SES) on CU traits in adolescents and found that CU traits increased as SES decreased only among adolescents with the homozygous long (l/l) genotype compared to those with the homozygous short (s/s) genotype. This suggests that there is a genetic risk for CU traits associated with a specific variant of the serotonin transporter gene only when children are raised in a disadvantaged environment.

Autonomic Nervous System

The autonomic nervous system (ANS) plays a significant role in emotion processing, and physiological responsivity is frequently measured using electrocardiogram or galvanic skin conductance recording. The ANS controls internal biological physiological responses to external stimuli and regulates automatic processes in the body such as maintenance of homeostasis (body temperature, blood pressure, breathing, and bodily fluids) and is comprised of the parasympathetic nervous system (PNS) and sympathetic nervous system (SNS). These systems enable nerves to trigger the body into "fight or flight" during stressful or emergency situations independently from the brain, while basic functions (e.g., heart rate and skin conductance) either increase (via SNS) or decrease (via PNS) in anticipation of the event. Therefore, the ANS provides useful insight into biological mechanisms that might underpin CD due to its management of reactivity to stressful situations (Berntson, Sarter, & Cacioppo, 2006). Several personality and neurobiological theories such as novelty-seeking behavior, fearlessness, and right hemisphere dysfunction (Ortiz & Raine, 2004) have been identified in the literature to account for the physiology behind CD. Males with CD present a generalized deficit in autonomic responsivity (Herpertz et al., 2005); indeed, a meta-analysis of 95 studies examining the association between skin conductance levels and antisocial behavior found that low heart rate and skin conductance response were consistently associated with psychopathy in adults and CP in children, likely reflecting aberrant emotion processing (Lorber, 2004). Children and adolescents with CD have slower resting heart rate levels than controls (INSERM_Collective_Expert_Reports, 2005; Ortiz & Raine, 2004). In addition, children with CP also show slower heart rates in response to aversive stimuli (Ortiz & Raine, 2004). Interestingly, psychological intervention has been shown to influence biological functioning in adults; for example, an increase in heart rate is observed following self-management of stress and emotion training (McCraty, Atkinson, Tomasino, Goelitz, & Mayrovitz, 1999). Furthermore, harsh parenting and a greater number of externalizing behaviors are associated with higher baseline sympathetic functioning in youths with CD (Bubier, Drabick, & Breiner, 2009; Ortiz & Raine, 2004). Skin conductance reactivity and startle reflexes/eye blinks of youths with adolescent- and childhood-onset CD were compared with no reported differences in fear conditioning between the two groups, despite the fact that both CD groups showed impaired differential fear conditioning relative to controls (Fairchild, Van Goozen, Stollery, & Goodyer, 2008).

Autonomic under-arousal, such as low heart rate and reduced skin conductance responsivity, has also been identified as a significant biomarker of CU traits in youths with CD (Blair, 1999), and some have hypothesized that this may predispose them to later psychopathy (Wang, Baker, Gao, Raine, & Lozano, 2012). Fung et al. (2005) found that a large sample of antisocial-prone youths with HCU traits had reduced anticipatory and skin conductance response to an aversive stimulus compared to those with LCU traits. These findings mirror those found in earlier studies on adults with high levels of psychopathic traits (Lynam et al., 2005). Furthermore, it has been suggested that reduced skin conductance response may be a partial driver for the sensation and novelty-seeking behaviors in children with CD/HCU traits (Raine, 2011).

As for heart rate, Anastassiou-Hadjicharalambous and Warden (2008) found that children with CD/HCU showed reduced heart rate compared to baseline when viewing emotional faces than children with CD/

LCU. However, this difference was not mirrored in self-report data; children with both CD/HCU and CD/LCU reported less empathic distress than controls, with no significant difference in self-report scores between CD groups. De Wied et al. (2012) replicated and extended these findings by showing that resting heart rate was not different between children with CP/HCU and those with CP/LCU traits, but resting respiratory sinus arrhythmia (variation in heart rate) was lower in those with CP/HCU traits compared to controls while witnessing another person in distress. This suggests that youths with CP/HCU traits exhibit reduced responsivity to emotion-provoking stimuli.

Sex differences in relation to ANS responsivity have been inconsistent. In a mixed gender sample of children with CD, hyporeactive skin conductance response was found in male adolescents with high levels of CU traits, but no association was reported for females (Isen et al., 2010). In contrast, in another study, males with CP were shown to have reduced skin conductance and heart rate, while females showed greater SCR responsivity compared to TD children (Beauchaine, Hong, & Marsh, 2008). Also, higher levels of aggression among males were found to be associated with reduced respiratory sinus arrhythmia or relative vagal tone, but no association was found among females (Beauchaine et al., 2008). Finally, in an all-female sample, Fairchild, Stobbe, Van Goozen, Calder, and Goodyer (2010) compared females with CD and TD females using differential autonomic fear conditioning. Results indicated that those with CD showed reduced skin conductance response to aversive unconditioned stimuli and deficient skin conductance discrimination with the conditioned stimuli, reflecting impaired fear conditioning. In sum, while it seems that males with CD consistently show reduced skin conductance compared to TD males, the evidence for females with CD is not as clear-cut.

Levels of Neurotransmitters

Atypical levels of neurotransmitters including serotonin, dopamine, noradrenaline, and oxytocin have all been identified in youths with CD (Matthys et al., 2013). Low serotonin functioning has been associated with increased levels of aggressive behaviour, particularly impulsive/reactive/hostile aggression (Buckholtz & Meyer-Lindenberg, 2008), but this association has not been consistent across studies (Duke, Begue, Bell, & Eisenlohr-Moul, 2013). Children with CD have reduced cortisol responsivity and altered serotonin, oxytocin, and noradrenaline neurotransmission, which can be framed in three domains: punishment processing, reward processing, and cognitive control (Matthys et al., 2013). Altered serotonin and noradrenaline transmission, along with reduced cortisol reactivity, suggests low punishment sensitivity, reflecting a failure for adolescents to anticipate punishment for their actions. Altered dopamine and low heart rate with respect to sensation seeking suggest reduced reward sensitivity. Finally, impaired functioning of the paralimbic system and executive functions suggest reduced cognitive control over behaviors (Matthys et al., 2013).

To our knowledge, no research has looked at the association between either age of onset of CD and neurotransmitters or sex and neurotransmitters. However, altered serotonin function may be implicated in the development of CU traits as suggested by the results of a recent study (Moul et al., 2013), which found that serum serotonin levels were significantly lower in males with CP/HCU traits than those with CP/LCU traits.

Neuropsychological Evidence

Impairments in executive functioning, including self-control, behavioral regulation, language comprehension, and emotion regulation, may lead to reduced behavioral disinhibition, increased risk taking, and abnormal reward/punishment processing (Seguin, Parent, Tremblay, & Zelazo, 2009). A meta-analysis of 39 studies with 4589 participants found that individuals with CD performed significantly worse on measures of executive function than controls (Morgan & Lilienfeld, 2000). This finding was replicated and extended in a more recent meta-analysis of

126 studies involving 14,786 participants, which reported that individuals with CD/ODD performed .54 standard deviations worse on executive functioning tests than controls (Ogilvie, Stewart, Chan, & Shum, 2011). There is also evidence that aggressive and nonaggressive symptoms of CD show different association patterns with measures of verbal abilities and executive function. Barker et al. (2011) found that poorer executive function and verbal abilities were associated with higher levels of physical aggressions symptoms, while theft was associated with higher scores on those cognitive domains. Although it was posited that youths with childhood-onset CD, but not those with adolescent-onset CD, were characterized by deficits in executive function compared to TD youths (Moffitt, 1993), a recent review of the literature concluded that there is "…little support for differences in executive function between childhood-onset and adolescence-onset forms of CD" (p. 931) and that in comparison to childhood-onset CD "…similar neurocognitive and psychophysiological impairments have been reported in adolescence-onset CD" (p. 932; Fairchild et al., 2013). Surprisingly, there is a paucity of research examining neuropsychological function in relation to CU traits in youths with CD, but one study observed a moderating role of CU traits on the association between verbal abilities and violent delinquency. Munoz, Frick, Kimonis, and Aucoin (2008) found that detained adolescents with HCU traits and high verbal abilities were more likely to report violent delinquency than those with LCU traits and high verbal abilities. A more recent study found that the highest levels of offending in youths were found in those with both high levels of CU traits and high IQ, demonstrating interactive effects between CU traits and cognitive ability (Hampton, Drabick, & Steinberg, 2014). The only study that has compared females with CD and TD females found that those with CD had lower IQ and performed more poorly on both visuospatial tasks and tasks of executive function (Pajer et al., 2008). However, methodological problems exist in this literature, with many studies using small samples that have not been objectively diagnosed with CD (Ogilvie et al., 2011).

Neuroimaging Correlates

Studies using functional (fMRI) and structural magnetic resonance imaging (sMRI) techniques have found evidence for abnormal brain function and structure in youths with CD in brain regions and networks implicated in affective and emotional face processing, empathy, morality, and decision-making (see Baker, Clanton, Rogers, & De Brito (2015) for a review). A key limitation of MRI studies on CD is that they are all cross-sectional. As a result, it is unclear the extent to which differences in brain function and structure observed in youths with CD are part of the etiology of this disorder, a consequence of the disorder and/or associated with environmental risk factors for CD, such as exposure to alcohol/drugs during pregnancy or childhood maltreatment.

Functional Neural Anomalies

Compared to TD youths, those with CD display atypical brain responses within frontal and temporal regions and limbic areas (Baker et al., 2015) in paradigms probing the processing of affective stimuli (Sterzer, Stadler, Krebs, Kleinschmidt, & Poustka, 2005), emotional faces (Passamonti et al., 2010), empathic response for others' pain (Decety, Michalska, Akitsuki, & Lahey, 2009), moral judgment making (Marsh et al., 2011), and decision-making (Rubia et al., 2009). One study identified subtle differences during conscious processing of facial expression of emotions in childhood- and adolescent-onset subtypes of CD (Passamonti et al., 2010). Compared to the TD group, both youths with childhood- and adolescent-onset CD showed reduced activation in the amygdala, ventromedial prefrontal cortex, insula, and orbitofrontal cortex (OFC) in response to angry faces, but youths with childhood-onset CD also exhibited reduced amygdala response to sad faces compared to TD youths and those with adolescent-onset CD, which might underlie their more persistent and severe clinical presentations compared to adolescent-onset CD (Passamonti et al., 2010).

In line with clinical and experimental data indicating that youths with CD/HCU and CD/

LCU have distinct affective profiles, recent fMRI work has revealed that higher levels of CU traits in adolescents with CP and CD are associated with reduced brain response in the amygdala during processing of fearful faces (Jones, Laurens, Herba, Barker, & Viding, 2009; Lozier, Cardinale, Van Meter, & Marsh, 2014; Marsh et al., 2008; Viding et al., 2012) and in the anterior cingulate cortex when seeing others in pain (Lockwood et al., 2013). For example, one study found that, compared to controls, youths with CP/LCU exhibited heightened amygdala response to fearful faces presented subliminally, whereas youths with CP/HCU showed lower amygdala response (Viding et al., 2012). The authors suggested that amygdala hypoactivity in CP/HCU adolescents could account for symptoms such as premeditated aggression, lack of empathy, and shallow affect, while amygdala hyperactivity in CP/LCU adolescents may explain these youths' propensity for threat-based reactive aggression and their difficulty in emotion regulation (Viding et al., 2012b).

As TD males and females show differences in brain development and brain functioning (Lenroot & Giedd, 2010), females with CD may present different impairments to males with CD. To date, only one fMRI study has compared females with CD to TD females and found that those with CD showed reduced medial orbitofrontal cortex and increased anterior insula activity when viewing emotional faces compared to healthy controls (Fairchild et al., 2014). These results differ to males with CD using the same task, where reduced amygdala, anterior insula, OFC, and anterior superior temporal cortex responses were observed, indicating that males have specific impairments in emotion processing (Passamonti et al., 2010).

In sum, these fMRI findings show that youths with CD display atypical brain responses in frontal and temporal areas, including the amygdala, during affective processing, and there is emerging evidence indicating that these brain responses are influenced by the age of onset of CD and levels of CU traits. More work is needed to examine potential sex differences in relation to brain response.

Structural Neuroimaging

Atypical neural responses in youths with CD might be partly underpinned by differences in brain structure and/or connectivity. Structural MRI studies have found that, compared to TD youths, those with CD exhibit reduced gray matter in brain areas associated with emotion processing and regulation, empathy, morality, and decision-making (Baker et al., 2015). One study that used voxel-based morphometry methods on a large sample of youths with CD ($n = 63$) found that male adolescents with CD exhibited reduced bilateral amygdala volume (extending into the insula) compared to TD youths (Fairchild et al., 2011). Furthermore, regression analyses demonstrated a negative correlation between number of conduct disorder symptoms and right insula volume in the CD group. When this group was divided into those with adolescent- and childhood-onset CD, it was found that while both groups showed reduced amygdala volume, only the adolescent-onset group showed reduced right insula volume relative to controls. These results are consistent with the fMRI evidence reviewed above, indicating reduced activity in those regions during processing of emotional facial expressions in youths with CD (Passamonti et al., 2010). Furthermore, youths with CD have been shown to have disrupted structural connectivity within white matter tracts connecting several cortical and subcortical regions, but the patterns of findings across studies have been inconsistent (Baker et al., 2015). Finally, a number of studies have looked at the association between CU traits and brain structure in CD/CP (De Brito & Hodgins, 2009; De Brito et al., 2011; Ermer, Cope, Nyalakanti, Calhoun, & Kiehl, 2013; Wallace et al., 2014). However, the directions of the association and the loci have been inconsistent across studies, with some finding a positive association (De Brito et al., 2009), no association (Fairchild et al., 2011), or negative association (Ermer et al., 2013; Wallace et al., 2014).

It is important to compare males and females, as research with TD adolescents has found that there are clear sex differences in the trajectory of

brain development (Lenroot & Giedd, 2010). Studies which have examined brain structure in females with CD have found that females have reduced gray matter volumes in temporal regions, which was stronger in females with CD than males with CD (Michalska, Decety, Zeffiro, & Lahey, 2015). A further study found that females with severe substance abuse and CP had 9.5% less overall gray matter volume compared to controls (Dalwani et al., 2015), with significant reductions in brain regions involved in inhibition, conflict processing, valuation of outcomes, decision-making, and emotion processing (Dalwani et al., 2015). However, a recent study did not observe group differences in white matter integrity between females with CD and TD females despite evidence of group differences in several white matter tracts between males with CD and TD males (Zhang et al., 2014). Further research is needed in order to pinpoint areas of structural abnormality in females with CD, as these may not directly mirror abnormalities observed in males with CD; a recent study found opposite pattern of gray matter differences between males and females with CD in the insula—a key region for empathy and decision-making (see Fairchild et al., 2013).

In brief, sMRI findings suggest that youths with CD show atypical brain structure and connectivity between regions central to emotion processing and regulation, empathy, morality, and decision-making. It must be noted, however, that the findings across studies are inconsistent, most probably due to variation in methods of analysis, sample characteristics such as different age ranges, and, for some studies, a failure to account for levels of CU traits (see Baker et al., 2015).

Treatment

The identification of aggressive behavior at a young age and the swift engagement of targeted services are key elements of positive behavioral improvement (Serketich & Dumas, 1996). Systematic reviews of randomized controlled trials have shown that early interventions have a positive impact on children with CD, although the relative efficacy of different approaches was unclear (Dretzke et al., 2009; Woolfenden, Williams, & Peat, 2001; Waddell, McEwan, Peters, Hua, & Garland, 2007). The heterogeneity of the disorder, coupled with high rates of comorbidity, means that a treatment option suitable for one child may not be effective for another (McCrory & Farmer, 2009). Frick (2001) postulates that a flexible approach to treatment is optimal, as the disorder is multifaceted and behavioral problems often affect different parts of an individual's life. Similarly, sustainability of interventions is vital; as children often begin to develop behavioral problems at a very young age, they require a consistent approach over a longer period of time in order to ameliorate their behavior (Kern et al., 2009). A large body of literature, commencing in the 1960s, has researched the efficacy of targeting and improving parenting styles. Dadds and Rhodes (2008) claim that a "coercive family process," an idea developed by Gerry Patterson in 1982 in which aggressive behavior is conditioned by parents and becomes self-sustaining, is one of the most eminent and important theories in the behavioral sciences. Modern research derived from neurobiological support also identifies drug treatments which target aggressive and impulsive behaviors (Gadow et al., 2014). Evidence-based approaches, including parent training, family therapy, individual and person-centered therapy, government-led programs, and pharmacological interventions, will be discussed below.

Parent Training and Parent-Child Interaction Therapy

One approach is to teach parents suitable parenting practices to effectively deal with their children's difficult behavior. Parent management training (PMT) is a common behavioral treatment for CD, where a therapist teaches parenting techniques, such as positive reinforcement and mild punishment (time-out), which are implemented at home (Kazdin, 2010). PMT usually spans over a period of several weeks and is conducted with

children up to the age of 12, and importantly, parents are instructed to monitor their own responses to their children rather than changing the environment of the child (McCrory & Farmer, 2009). In a sample of children from Norway, externalizing behaviors decreased and parent and teacher behavior ratings increased, documenting the effectiveness of PMT (Ogden & Hagen, 2008). Additionally, parent-child interaction therapy (PCIT) has been shown to reduce defiant behaviors among toddlers (Eyberg, Boggs, & Algina, 1995). During PCIT, parents and children play while a therapist gives guidance over a headset in a different room on how to appropriately converse with their child. Evaluation has shown effectiveness in reduction of behavioral problems, decrease in parental stress, and lower levels of noncompliance up to 6 weeks after treatment (Eisenstadt, Eyberg, McNeil, Newcomb, & Funderburk, 1993). Parenting behaviors such as more parental warmth and praise have been found to be associated with lower levels of CU traits (Pasalich, Dadds, Hawes, & Brennan, 2011). Early attention to eye contact with attachment figures may be an important cue to parents. In a sample of young children (M age = 8 years), youths with HCU rejected eye gaze regardless of the mother's attempts to show affection, and higher levels of psychopathic traits in the paternal caregiver were associated with low levels of eye gaze in the child (Dadds et al., 2014).

Family Therapy

Systematic approaches have proved successful, particularly "multisystemic therapy" (MST), an intensive, full-family intervention approach that addresses all aspects of home life and identifies areas of improvement of young offenders (Henggeler, 2002; Henggeler et al. 1992). Juvenile delinquents who received family and parenting interventions have been found to be less likely to reoffend and spend less time incarcerated than those who do not receive this treatment (Woolfenden, Williams, & Peat, 2002). Functional family therapy (FFT) is another full-family strategy which aims to improve levels of support and healthy communication between family members (Parsons & Alexander, 1973). While some authors claim that these interventions lead to robust improvements in child behavior (Weisz & Kazdin, 2010), others claim that improvement is mixed (Woolfenden et al., 2002). Thus, the labor-intensive nature of the interventions, along with mixed evidence that the interventions improve family functioning and peer relations, means that the efficacy of these treatments should be carefully considered (Woolfenden et al., 2002).

Individual and Person-Centered Therapy

There have been several cognitive behavioral therapies (CBT) designed to treat adolescents with CD directly. One such approach is cognitive problem-solving skills training (PSST), which is designed to help adolescents process and respond to situations through role play with a therapist who guides the adolescent on how to appropriately react and respond to real-life situations (Kazdin, 2010). The main goal of this approach is to deter impulsive responses and make appropriate decisions in social settings. Other individual therapies target emotion regulation and management of noncompliance, anger, and frustration (Weisz & Kazdin, 2010). Heterogeneity is also important to consider when treating children with CD, and children with CD/HCU traits are oftentimes more difficult to treat due to their resistance and evidence of little change (Hawes & Dadds, 2005). One study in children aged 7–12 years reported that, compared to children with CP/LCU traits, those with CP/HCU traits did not respond as well to behavioral therapy, which involved a series of activities led by counselors where a point system was implemented to reward good behavior (Waschbusch, Carrey, Willoughby, King, & Andrade, 2007). Frick and Marsee (2006) posit that these different responses to the same treatment may be due to the distinct developmental processes which underlie each subgroup. However, another study showed that when very young children (mean age 3.8 years) with CP/HCU traits were given such therapy, their behavioral issues were reduced, reinforcing the view that early interventions for children with CP/

HCU traits might be an important factor for successful outcomes (Kimonis et al., 2014). To address variability in responsiveness to treatment, emotion recognition training has been adapted for children with behavioral difficulties and has been shown to improve CP and affective empathy in children with high levels of CU traits (Dadds, Cauchi, Wimalaweera, Hawes, & Brennan, 2012). Finally, a recent systematic review focusing on behavioural therapy, emotion recognition training and multimodal interventions noted that, while children with CP/HCU traits have poorer premorbid functioning at the beginning of the intervention and can still exhibit with higher levels of antisocial behaviour than other youths at the end of the intervention, CU traits and antisocial behaviour in those children can be reduced (Wilkinson, Waller, & Viding, 2016).

Government-Led Programs

Several initiatives have been developed by government systems which have been instigated in schools with the aim to reduce CP in young children. The "Incredible Years Classroom Dinosaur Programme" was developed and adapted in the UK to improve behavior in the classroom, using methods such as videotape modelling, reinforcement of positive behavior, and role play (Webster-Stratton & Reid, 2004). In the USA, Head Start schools have been developed to help disadvantaged children improve social skills and emotional self-regulation; children from these schools exhibit fewer behavioral problems than TD children (Webster-Stratton, Jamila Reid, & Stoolmiller, 2008). Crucially, this type of intensive approach seems to have long-term benefits as these effects were still evident 1 year after training had ended (Webster-Stratton, Reid, & Hammond, 2001).

Pharmacological Interventions

Pharmacological treatments are not prescribed for CD, but many adolescents with CD receive medication for comorbid disorders such as ADHD. One study on adolescents with CD, 65% of whom had comorbid ADHD, looked at the efficacy of methylphenidate, a commonly prescribed treatment for ADHD. The study concluded that antisocial behaviors (rated by parents or teachers) were reduced after treatment, even when ADHD severity was controlled for in the analysis (Kratochvil et al., 2002). Methylphenidate may also improve facial emotion recognition in adolescents (von Morgenstern, Becker, & Sinzig, 2014), a skill that is impaired in adolescents with CD (Fairchild, Van Goozen, Calder, Stollery, & Goodyer, 2009). A more recent study showed that risperidone, when added to stimulant medication for adolescents with CD/ODD and ADHD, ameliorated levels of ODD, but not CD symptoms (Gadow et al., 2014).

Since the 1970s, lithium has been used to treat manic depression, as it reduces symptoms of mania and hyper-aggressiveness (Sheard, 1975). One study has looked at lithium as a potential treatment for children and adolescents hospitalized for CD and severe aggression and found that short-term lithium treatment reduced aggressive behaviors, albeit with side effects including headaches and urinary frequency (Malone et al., 2000), which means that lithium is not a common treatment for CD. More recently, based on several lines of evidence, Netherton and Schatte (2011) recommend that oxytocin (OT) should be examined as a potential treatment for CD, as it may reduce aggression, increase trust and social skills, and make adolescents with CD less likely to misidentify emotions. Reasoning for this includes the following: low levels of OT are associated with aggression (for review, see Heinrichs & Domes, 2008), men are better at reading others' emotions after OT administration (Domes, Heinrichs, Michel, Berger, & Herpertz, 2007), and finally, participants are more likely to remember previously seen happy faces than angry and neutral faces after OT administration (Guastella, Mitchell, & Dadds, 2008) and are less likely to mislabel positive expressions as negative (Di Simplicio, Massey-Chase, Cowen, & Harmer, 2008). It has been suggested that it would be of great clinical significance if one could increase OT levels without medicinal intervention. An interesting avenue for future studies to pursue involves administering

OT to mothers (whose children are at risk of developing CD) during pregnancy, which may increase bonding behaviors and increase the child's OT levels naturally (Netherton & Schaffe, 2011).

Prognosis and Outcome

Mental health disorders in adulthood are a continuation of juvenile mental health disorders (Copeland, Shanahan, Costello, & Angold, 2009). With regard to CD, there is evidence of homotypic continuity with several prospective, longitudinal, and cross-sectional studies of large population cohorts reporting that the adult syndrome indexed by ASPD begins in childhood and persists across the life-span (Kim-Cohen et al., 2003; see De Brito & Hodgins, 2009, for a review). Thus, the DSM requirement that a diagnosis of ASPD is given only if CD was present prior to age 15 is supported by a substantial body of research. As noted before, the younger the age of onset and the higher the number of symptoms, the greater the likelihood that CD develops into ASPD in adulthood (Lahey et al., 2005). While, by definition, all adults with a diagnosis of ASPD have a childhood history of CD, it is important to emphasize that only about 50% of children with CD develop ASPD (Simonoff et al., 2004). There is also evidence of heterotypic continuity (Rowe et al., 2010); for example, a longitudinal study spanning several decades showed that between 25% and 60% of adults diagnosed with *any* mental health disorder also had CD or ODD before the age of 15. Both men and women with a history of CD were more likely to have any axis I disorder, axis II disorder, any alcohol use disorder, and any drug abuse disorder than those without a diagnosis of CD (Kim-Cohen et al., 2003). The heterogeneity associated with CD means that the different subtypes can have very different prognoses.

Those with childhood-onset CD are more likely to offend in adulthood and spend a larger amount of time in prison than those with adolescent-onset CD (Moffitt, Caspi, Harrington, & Milne, 2002). Adults who were diagnosed with childhood-onset CD are more likely to smoke and have poorer health in adulthood, including increased hospital visits, a greater chance of developing sexually transmitted disorders, and a higher chance of developing substance use disorder and personality disorders compared to those without an early childhood diagnosis of CD (Odgers et al., 2007; Moffitt, 2003). While those with adolescent-onset CD have better predicted adult outcomes, their education, work, health, and family life are relatively unimpaired, and their adult prognosis also includes substance abuse and minor and/or undetected crimes (Odgers et al., 2007; Nagin, Farrington, & Moffitt, 1995).

A comprehensive review of 118 studies reported that CU traits are predictive of later aggression, antisocial behavior, and psychopathic tendencies (Frick et al., 2014a). Children and adolescents with CD/HCU traits are less responsive to treatment and interventions than those with CD/LCU traits (Hawes & Dadds, 2005). Furthermore, those with HCU traits exhibit more severe and stable antisocial behavior (Frick, Stickle, Dandreaux, Farrell, & Kimonis, 2005) and are more likely to reoffend with violent crimes in a 2-year period after being released from jail (Vincent, Vitacco, Grisso, & Corrado, 2003). One study found that parent- and teacher-rated levels of CU traits in 500 7-year-old males predicted criminal behavior at age 25, despite controlling for CD, ADHD, and ODD (Byrd, Loeber, & Pardini, 2012). Lynam et al. (2007) showed that the presence of psychopathic traits at 13 years was a moderate predictor of psychopathy at 25 years; therefore, early intervention and treatment should be a priority.

The available data suggest that females with CD show a similar prognosis to males with CD (Odgers et al., 2007; Bardone et al., 1998, meta-analysis). For example, Bardone et al. (1998) found that females who had CD at 15 years were more likely to have poorer general health, medical problems, drug and tobacco dependence, and sexually transmitted diseases and become pregnant by 21 years old than females without CD. Females with CD are also more likely to

have children at a younger age; 72% of those with childhood-onset CD and 61% of those with adolescent onset CD had children at age 32, compared to 50% of control women who had children by this age (Odgers et al., 2008). A longitudinal study found that females with childhood-onset CD were more likely than those with adolescent-onset CD to continue to show antisocial behavior and suffer from mental health, physical, and economic problems (Odgers et al., 2008). In contrast to females with childhood-onset CD, females with adolescent-onset CD did not show mental and physical health problems and were less likely to continue their early pattern of antisocial behavior into adulthood, although they also suffered from economic problems (an outcome that may be influenced by related factors such as teenage pregnancy) (Odgers et al., 2008).

Conclusion

There is no single factor that causes CD to develop. This review has discussed various individual and environmental risk factors, concluding that while no one factor is more salient than any others, all play a role and interact with each other in the development of CD. Given the overwhelming evidence of the heterogeneous nature of the disorder, research on etiology and delineation of developmental pathways and subtypes and their associated risk factors should be a priority to develop more tailored and efficient treatments (Frick & Viding, 2009). Large-scale studies, such as the FemNAT-CD project (www.femnat-cd.eu), which uses multimodal approaches and multiple levels of analyses to identify biomarkers and environmental factors implicated in the development and persistence of CD in males and females, are an important avenue for future research. Fifty percent of children with CD continue onto lifetime offending patterns, which later develop into severe delinquency (Simonoff et al., 2004). While it is important to understand how the interplay between individual and environmental risk factors contribute to the onset and the persistence of CD in some youths and its escalation into adult ASPD, there is also a pressing need to understand the mechanisms that contribute to desistence in 50% of cases, as protective factors and insight into resilience might prove equally important in developing prevention and intervention efforts. Since these behaviors are detectable early in life, children with CD, their family, and society at large would benefit while they are more susceptible to intervention strategies (Frick, 2001).

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Pyromania

27

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History of Pyromania in the DSM

Pyromania's inclusion as a mental disorder in the *Diagnostic and Statistical Manual of Mental Disorders* (DSM) has varied historically. Pyromania appeared in the first edition of the DSM (American Psychiatric Association [APA], 1952) as a supplementary term within the appendices that could be added to a diagnosis to further explain the presentation of a patient. However, no details were offered about the term, and no indication was given regarding whether it could be used with children or adolescents in addition to adults. Despite its inclusion in DSM-I, pyromania did not receive any mention in the second edition of the manual (DSM-II; APA, 1962). It was re-included in the third editions as an *impulse control not elsewhere classified* (DSM-III; DSM-III-R; APA, 1980, 1987) and remained classified as such in both of the fourth editions (DSM-IV; DSM-IV-TR; APA, 1994, 2000). Interestingly, throughout all four editions, no information was offered regarding the prevalence of the disorder other than to specify pyromania as being "apparently rare" but more prevalent among males. Further, very little was said of the course of the disorder other than that "fire-setting incidents are episodic and may wax and wane in frequency" (DSM-IV; DSM-IV-TR; APA 1994, 2000).

The third editions of the DSM explicitly indicated that pyromania could onset in childhood. More specifically, DSM-III and DSM-III-R stated that onset is usually in childhood but that adolescent- or adult-onset pyromania is typically more deliberately destructive in nature. However, later revisions were less clear about the possibility of pyromania in childhood. The fourth editions of the DSM (DSM-IV and DSM-IV-TR) indicated that evidence was insufficient to establish age of onset of pyromania. They also reported that the relationship between firesetting behavior in childhood and subsequent adult pyromania was not documented. No information was provided in DSM-III through DSM-IV-TR regarding the longitudinal course of the disorder.

The diagnostic criteria for pyromania underwent several changes across the different editions and revisions of the DSM. DSM-III (APA, 1980) required a recurrent "failure to resist impulses to set fires" (p. 295), while the revised text of the third edition (p. 326) and all subsequent editions referred to "deliberate and purposeful fire setting on more than one occasion." All versions referred to tension prior to setting fire with DSM-III describing an "increasing sense of tension before setting the fire" (p. 295) and subsequent editions broadening this slightly to "tension or affective

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arousal before the act." DSM-III did not specifically refer to fire fascination within the diagnostic criteria, although it did refer to an "intense fascination with setting fires and seeing them burn" (p. 294) as an essential feature of the disorder in the overview. Later editions, however, included fire fascination within the diagnostic criteria. The most detailed criterion description regarding fire fascination was included in DSM-III-R (APA, 1987, p. 326) as: "Fascination with, interest in, curiosity about, or attraction to fire and its situational context or associated characteristics (e.g., paraphernalia, uses, consequences, exposure to fires)." However, the "associated characteristics" and "exposure to fires" elements were dropped in subsequent editions. The DSM-III described experiences of pleasure, gratification, or release only with regard to the committal of a firesetting act. Subsequent editions allowed for these affective responses to be considered when the individual is witnessing or participating in the aftermath of firesetting behavior. All editions of the DSM included extensive exclusionary criteria relating to (1) the motivations for firesetting or (2) alternative diagnoses that might better explain firesetting behavior. Motives that excluded a pyromania diagnosis included monetary gain; sociopolitical ideology (DSM-III, DSM-III-R, DSM-IV, DSM-IV-TR), concealing criminal activity; expressing anger or vengeance, for improvement of living circumstances, as a response to delusions, hallucinations, or intoxication (DSM-III-R, DSM-IV, DSM-IV-TR); or firesetting as a result of impaired judgment such as in dementia, intellectual disability, or substance intoxication (DSM-IV, DSM-IV-TR). DSM-III specified that firesetting should not be due to "an organic mental disorder, schizophrenia, antisocial personality disorder, or conduct disorder." These exclusions were absent from the diagnostic criteria in DSM-III-R. Both versions of the fourth edition of the DSM specified that firesetting should not be "better accounted for by conduct disorder, a manic episode, or antisocial personality disorder." Across all editions of the DSM prior to DSM-5, the diagnostic criteria made it very difficult for clinicians to confidently diagnose pyromania in children or in adults.

Presentation of the Current Diagnostic Criteria in DSM-5

DSM-5 (APA, 2013) classifies pyromania among the *disruptive*, *impulse control*, *and conduct disorders*. The diagnostic criteria for pyromania did not change substantively in the fifth edition compared with those outlined in the DSM-IV and DSM-IV-TR. The only difference is the labeling of the examples given in criterion E (see Table 27.1). Despite reordering of sections, the DSM-5 does not offer the consulting clinician a considerably more detailed picture of the disorder than previous editions, with the exception of slightly more information on prevalence.

DSM-5 offers little additional information over previous editions to clinicians working with children and young people who have set fires. One difference of DSM-5 compared to earlier editions is that disorders have been organized to reflect developmental and life span considerations. Thus, DSM-5 begins with disorders thought to develop early in life, following on to those which onset in adolescence and early adulthood, and concludes with those which are thought to onset in adulthood and later life. Within this

Table 27.1 DSM-5 diagnostic criteria for pyromania

Diagnostic criteria	
A.	Deliberate and purposeful firesetting on more than one occasion
B.	Tension or affective arousal before the act
C.	Fascination with, interest in, curiosity about, or attraction to fire and its situational contexts (e.g., paraphernalia, uses, and consequences)
D.	Pleasure, gratification, or relief when setting fires or when witnessing or participating in their aftermath
E.	The firesetting is not done for monetary gain, as an expression of sociopolitical ideology, to conceal, criminal activity, to express anger or vengeance, to improve one's living circumstances, in response to a delusion or hallucination, or as a result of impaired judgment (e.g., in major neurocognitive disorder, intellectual disability [intellectual developmental disorder], substance intoxication)
F.	The firesetting is not better explained by conduct disorder, a manic episode, or antisocial personality disorder

structure, DSM-5 asserts that, despite the fact that a large proportion of individuals arrested for arson are under 18 years of age, "pyromania in childhood appears to be rare" (p. 477). This appears to suggest that this is a disorder predominantly diagnosed in adulthood. However, there is a significant lack of literature referring to childhood pyromania; it is therefore possible that this assertion is due to the poor evidence base rather than being derived from a body of adequately controlled empirical literature.

It is also possible that the omission of pyromania from the modern research narrative of firesetting in childhood and adolescence is due, in part, to the potentially stigmatizing nature of the label. DSM-5 states that childhood firesetting may occur as part of developmental experimentation but offers no opinion on the relationship between childhood firesetting and adult pyromania. Further, deliberate firesetting is also described as being symptomatic of other childhood disorders within DSM-5. For example, "deliberate firesetting with the intention of causing damage" is listed as a clinical feature of conduct disorder (p.470, DSM-5; APA, 2013). Conduct disorder is classified as part of the same groups of disorders as pyromania within DSM-5. Conduct disorder is considered to onset either during childhood (prior to age 10) or during adolescence (post age 10) and is characterized by a repetitive and persistent pattern of behavior where the rights of others and/or societal norms are violated. Key clinical features of conduct disorder include aggression, destruction of property (including firesetting), deceitfulness or theft, and serious violations of rules. Thus, it is possible that many children who engage in repetitive firesetting may be more likely to attract a diagnosis of conduct disorder than one of pyromania. Point "F" of the diagnostic criteria for pyromania provides some support for this by stating that if firesetting is better explained by conduct disorder, a manic episode, or antisocial personality disorder, then this should be given over a diagnosis of pyromania.

Overall, despite adjustments to the DSM criteria for pyromania over successive editions, the latest criteria in DSM-5 continues to make it extremely unlikely that an individual will be diagnosed with the disorder given that for most individuals presenting with firesetting behavior, differential diagnosis will suggest other causes of intentional firesetting or other mental disorders as better explanations of firesetting. The potential for research examining the developmental course of pyromania is consequently hamstrung by these limitations.

Current Research Findings on Pyromania

Relative to some earlier uses of the term pyromania, the current diagnostic criteria in DSM-5 leaves the construct so narrowly defined that it is very rarely diagnosed. Other reviews or reference guides accommodate the paucity of research or clinical evidence surrounding pyromania by drawing on sources that adopt different definitions of pyromania or on studies that examine fire interest more broadly. However, synthesizing such evidence runs the risk of muddying the waters by equating phenomena that should not be considered equivalent or by pathologizing behaviors that would not fall under a DSM-5 diagnosis of pyromania. In this chapter, to improve clarity we will only examine research that explicitly refers to pyromania as defined by the DSM-III or later. In preparing this research review, we conducted thorough searches for evidence which yielded an extremely small volume of empirical findings on pyromania in young people. For example, we carried out a search on Scopus for articles using the search terms pyromani* and child* or adolesc* within title, abstract, and keywords and DSM* or "International Classification of Diseases" in all fields. This search yielded only 16 matches, many of which were not particularly useful (e.g., short discussions of DSM-defined pyromania). As a result, in the following sections, we draw on any identified relevant literature relating to children and adolescents using database and supplemental search strategies but also incorporate (and are sometimes dependent on) findings from research on adults with pyromania. We state the age ranges of participants in the cited studies, where this information is available.

Prevalence

Latest available statistics show that between 2007 and 2011, US fire departments received reports of approximately 282,600 deliberate fires each year which were annually responsible for 1360 casualties, 420 deaths, and $1.3 billion costs in property damage (Campbell, 2014). Children and adolescents account for a large percentage of arrests for arson. According the Federal Bureau of Investigation (2014), 20% of individuals arrested for arson in the USA were under 15, and 35% were under 18. However, while deliberate firesetting by young people and adults is clearly a huge public safety issue, it remains unclear how much firesetting is accounted for by individuals with pyromania.

Because diagnoses of pyromania are extremely rare, very few studies have applied DSM-III (or later) criteria and provide information about the prevalence of pyromania. Those that do tend to examine adult convenience samples and do not necessarily yield consistent or comparable findings. We first outline findings from studies using DSM-IV criteria or later before discussing research using criteria based on DSM-III and DSM-III-R. Lindberg, Holi, Tani, and Virkkunen (2005) found that only three individuals (3.3%) from a sample of 90 incarcerated male repeat firesetters, aged over 16 years, fully met the criteria for pyromania. A further nine individuals fulfilled all inclusion criteria except substance intoxication. Examining a sample of 25 remanded female prisoners (ages not reported) with a history of self-mutilation and firesetting, Coid, Wilkins, and Coid (1999) found that 14 (56%) received a diagnosis of pyromania. Dickens et al. (2007) examined a sample of male and female adults (aged 18–77) referred to a forensic psychiatry service for arson. They reported that 31 of 129 males (24%) met the DSM-IV criteria for pyromania. They do not state how many, if any, females met these criteria but report that "several" of a total of 38 females were preoccupied with fire. None of these studies comment on whether symptoms of pyromania were present in the identified individuals during childhood or adolescence.

Grant, Levine, Kim, and Potenza (2005) reported lifetime prevalence of pyromania among 204 male and female adult psychiatric inpatients (aged 18–83) of 5.9% (majority male; Grant & Kim, 2007) using the Minnesota Impulsive Disorders Interview (MIDI) and a structured clinical interview. Current prevalence was 3.4%. Grant, Williams, and Potenza (2007), again using the MIDI, found evidence for pyromania among 12.5% of 56 female adolescent psychiatric inpatients but no evidence of the disorder among 46 adolescent male psychiatric inpatients (combined sample age range was 13–18). This finding with an adolescent sample is interesting given that DSM-IV asserts that pyromania occurs "much more often in males" (p. 477).

Using the MIDI with a larger sample of adults examined in a psychiatric emergency ward, Adam, Richoux, and Lejoyeux (2008) reported two individuals (1%) with pyromania. Evidence of pyromania using the MIDI appears rare when nonpsychiatric samples are included. Black et al. (2015) examined the impulse control disorders of 95 pathological gambling probands, 91 controls, and 1075 first-degree relatives, all aged 18 years or older. They did not report a single case of pyromania. Odlaug and Grant (2010) found slightly more evidence of pyromania using a self-administered version of the MIDI with a sample of 791 college students (aged 17–24). The responses of eight individuals (1%) were indicative of pyromania. Rates of pyromania did not differ significantly based on gender.

Using DSM-III or DSM III-R criteria for pyromania, which had fewer exclusions than with DSM-IV and beyond, three studies found no evidence of pyromania among small samples (*n*s 26–29) of male and female firesetters aged 17 or above (Harmon, Rosner, & Wiederlight, 1985; Koson & Dvoskin, 1982; Leong, 1992), and a fourth study only found one individual meeting the criteria for pyromania among a sample of 243 mentally disordered adult male firesetters (Rice & Harris, 1991). Among a sample of 45 young male firesetters who were 21 or younger at the time of their first firesetting offense, and who were referred to pretrial psychiatric examination, Repo and Virkkunen (1997) identified nine (20%) individuals with pyromania.

Taken together it is extremely difficult to be confident about the true prevalence of pyromania among children and adolescents as well as among adults. The studies above are all limited in terms of sample size and rely on convenience samples, and some offer wildly different indications of the number of individuals with pyromania across different samples. Some of the differences across studies may well be due to different applications of diagnostic criteria for pyromania. The lack of studies on which to base the prevalence of pyromania in young people may reflect a genuine absence or rarity of the disorder in that population. However, it may also reflect that symptoms of pyromania may be more easily subsumed within a conduct disorder diagnosis or that concerns over the stigmatizing effects of a pyromania diagnosis discourages its use. In order to establish the true prevalence rates of pyromania, prospective cohort studies should be carried out.

Etiology

It is not clear to what degree we can generalize the results of studies using small samples to individuals with pyromania more broadly. With these caveats in mind, we present the evidence on the etiology and other elements of pyromania based on the available literature.

There is little or no empirical data on the construct of pyromania in childhood and adolescence (MacKay, Feldberg, Ward, & Marton, 2012). Grant and Kim (2007) combined samples of adults and adolescents with pyromania from two studies (Grant et al., 2005; Grant et al., 2007) along with a further two individuals with lifetime pyromania and examined the clinical characteristics and comorbidity of the disorder. We draw heavily on this study to underpin our conclusions throughout this chapter as it is the only study that examines a "pure" sample of individuals with pyromania in sufficient detail to inform the clinician as to what *may* be typical for individuals with this diagnosis. DSM-5 states that there is no sufficient evidence to determine a typical age of onset for pyromania. Grant and Kim's (2007) sample included seven adolescents (all females) and 14 adults (three females). For these 21 participants, mean age of onset was 18.1 (standard deviation [SD] = 5.8; range 12–33) years. This suggests (assuming a normal distribution) that the onset of pyromania symptoms occurred prior to the age of 18 for approximately half the sample. Coid et al. reported a mean age of onset of firesetting behavior of 16.5 (SD = 2.6) years for their sample of females with pyromania (and self-mutilation). This was younger than their non-pyromaniac female firesetters (M = 20.3 years; SD = 9 years), though this difference was nonsignificant. This finding also refers to the onset of firesetting behavior rather than the onset of pyromania symptoms per se.

To our knowledge, there is no empirical research that demonstrates clear evidence for causal mechanisms underpinning pyromania. Both Grant and Kim (2007) and Coid et al. (1999) suggest that the developmental backgrounds of a sizeable number of their participants with pyromania were characterized by physical abuse, sexual abuse, or other deprivation. However it is unclear to what degree the observed levels differ from other individuals in a psychiatric setting. Various single-factor (e.g., psychoanalytical, social learning, biological) and multifactor theories have been proposed to explain the causes of firesetting (see Gannon, Ó Ciardha, Doley, & Alleyne, 2012; Gannon & Pina, 2010). With the exception of psychoanalytical theory, none of these theories pay specific attention to pyromania. Psychoanalytical theory has not demonstrated empirical adequacy and additionally does not conceptualize pyromania consistent with the DSM classification.

Symptom Presentation

The defining clinical features of pyromania in DSM-5 are multiple occasions of deliberate firesetting combined with tension or arousal prior to setting the fire and pleasure gratification or relief following setting the fire. Furthermore, there must be evidence of interest, curiosity, or attraction toward fire. While the International Classification of Diseases (ICD-10) allows for

attempted firesetting within its criteria of pathological firesetting or pyromania, the DSM-5 assumes the deliberate and purposeful setting of an actual fire. Grant and Kim (2007) presented the clinical characteristics of their subset of individuals with pyromania. The frequency of firesetting behavior was one fire every 2–16 weeks (*M* = 5.9, SD = 3.8 weeks). Many of the fires set by their sample may not necessarily have met legal definitions of arson. Two thirds of their sample engaged in planning (including buying materials), while the remaining third described their fires as generally unplanned.

While all of the participants in Grant and Kim's (2007) study reported feeling a "rush" while watching or setting fires, none reported sexual excitement. Despite early research and theory on pyromania focusing on sexual motivations for firesetting, especially among researchers from a psychoanalytic tradition, there is very little evidence for this as a common motive (see Ó Ciardha, 2016). While it appears that some firesetters do have a sexual component to their firesetting behavior (e.g., Kocsis & Cooksey, 2002), and that a "significant minority of arsonists show greater sexual arousal to firesetting themes than do controls subjects" (Harris, Rice, Quinsey, Chaplin, & Earls, 1992; p. 508), it is not clear from the current literature how many of these individuals would meet the DSM-5 diagnostic criteria for pyromania.

In Grant and Kim's (2007) study, the majority (86%) of individuals with pyromania reported urges to set fire, which they could control 64.5% of the time (SD = 15.5%, range = 33.3%–90.0%). The most common reported triggers for firesetting were stress, boredom, feelings of inadequacy, and interpersonal conflict. The vast majority (91%) of individuals reported feeling severe distress after setting fires and 31% reported considering suicide to control their firesetting. This was despite the fact that many also reported pleasure and/or relief when setting the fires. A third of those sampled by Grant and Kim (2007) reported that time spent watching fires or planning and engaging in firesetting caused significant social or occupational impairment. Importantly, Grant and Kim (2007) did not identify any significant differences in the clinical characteristics of adolescents with pyromania compared to adults. However, it is clear that larger samples might have greater statistical power to examine whether younger individuals with pyromania exhibit unique symptomology.

Associated Impairments and Comorbidities

DSM-5 diagnostic criteria indicate that firesetting should not be better accounted for by conduct disorder, a manic episode, or antisocial personality disorder. Furthermore, for a pyromania diagnosis, firesetting must not be conducted in response to a delusion or hallucination or be as a result of impaired judgment, due, for example, to dementia, intellectual disability, or substance intoxication. As a result, research involving individuals with strictly defined pyromania may give a different view of comorbidity than studies that look at less narrowly defined firesetting behavior. DSM-5 is the first edition of the DSM to have included information on comorbidity. It states that there "appears to be a high co-occurrence of substance use disorders, gambling disorder, depressive and bipolar disorders, and other disruptive, impulse-control, and conduct disorders with pyromania" (p. 477). It is not clear what evidence there is for some of the assertions that these disorders are often comorbid. For example, the fact that firesetting should not be better explained by conduct disorder, coupled with the fact that firesetting should not be carried out to express anger or vengeance, would suggest that the consulting clinician would rarely diagnose comorbid pyromania and conduct disorder (MacKay, et al., 2012). According to Grant and Kim (2007), the most common lifetime comorbidities among their sample of individuals with pyromania were impulse control disorders (66.7%), affective disorders (61.9%), anxiety disorders (33%), and substance use disorders (33%). As was the case with the clinical presentation of individuals with pyromania, they reported no significant differences in rates of comorbid disorders between adolescents and adults.

Grant and Kim (2007) excluded individuals from their study if they met the criteria for conduct disorder, bipolar disorder, psychotic disorders, or developmental disorders. As a result, their study may have excluded individuals with pyromania and comorbid disorders. Grant and Kim (2007) did not exclude individuals meeting the criteria for antisocial personality disorder (ASPD). However, they reported that no one met the criteria for a diagnosis of ASPD. Several studies report that ASPD is associated with firesetters and firesetting behavior (Ducat, Ogloff, & McEwan, 2013; Lindberg, et al., 2005; Rix, 1994). Furthermore, Coid et al. (1999) found high levels of ASPD in their sample of females with pyromania and a history of self-mutilation. In addition to a lack of ASPD in Grant and Kim's (2007) study, only two participants (9.5%) met the criteria for borderline personality disorder, which is also associated with firesetting behavior (see Ó Ciardha et al., 2015). This apparent absence of personality disorders in their sample raises two possibilities—either (1) the sample in Grant and Kim's (2007) study is not representative of the range of comorbid disorders suffered by individuals with pyromania or (2) pyromania may not have the same relationship with psychopathology as is found among individuals where factors other than pyromania cause firesetting behavior. This illustrates the points made earlier about treating the results of such small-scale studies as Grant and Kim's (2007) with caution and also about the risk of muddying the waters by conflating pyromania and firesetting more broadly in order to fill the vacuum left by the paucity of research examining pyromania explicitly.

Developmental Course

While firesetting behavior is a major problem in children and adolescents, DSM-5 states that there is a lack of evidence regarding the relationship between this behavior and adult pyromania. It also states that while firesetting incidents are episodic and may wax and wane, the longitudinal course of the disorder is unknown. Grant and Kim (2007) reported that for their five participants who did not meet the criteria for a current pyromania diagnosis but who did have lifetime pyromania, mean duration of pyromania symptoms was 8.2 (SD = 0.8; range 7–9) years. Interestingly, each of these individuals reported that their firesetting symptoms had ceased when they began another impulsive or compulsive behavior. As mentioned, frequency of firesetting behavior for those experiencing active pyromania was once every 2–16 weeks. Only two individuals (10%) in Grant and Kim's study had been arrested for their firesetting behavior, suggesting that large numbers of individuals with pyromania may not come to the attention of the criminal justice system. The majority reported feeling urges to set fires that were often, but not always, controllable. Looking at individuals apprehended and assessed as a result of firesetting, Repo and Virkkunen (1997) found that, relative to other young male firesetters, individuals with a diagnosis of pyromania had the highest rates of both firesetting and non-firesetting recidivism. This appears consistent with Grant and Kim's (2007) finding that over the course of the disorder, frequency and intensity of firesetting behavior increases. However, Repo and Virkkunen (1997) also found that 65% of their sample of male adolescent firesetters had a history of conduct disorder and concluded that, among this population, firesetting is most likely associated with childhood-onset conduct disorder.

Treatment

The rarity of diagnoses of pyromania in adults or children is an obvious barrier to the systematic evaluation of treatment efficacy for this disorder. While other forms of impulse control disorders have a limited but increasing evidence base on which to judge the effectiveness of pharmacological or psychotherapeutic interventions, no randomized controlled clinical trials have been conducted for interventions designed to treat pyromania in either adults or children. Pharmacological interventions that have been used in individual cases with apparent partial or

complete remission of symptoms (though not always maintained after discontinuation of medication) include escitalopram, sertraline, fluoxetine (SSRIs), topiramate (antiepileptic), lithium (mood stabilizer), and olanzapine (neuroleptic) combined with sodium valproate (antiepileptic; Grant & Odlaug, 2011; Grant & Kim, 2007; Parks et al., 2005). Grant and Kim (2007) point out, however, that among their sample, the majority of those treated with pharmacological interventions did not respond to treatment (despite some of the same medications apparently leading to positive results in other cases). Interventions included fluoxetine, valproic acid, lithium, sertraline, olanzapine, escitalopram, citalopram, and clonazepam. It is worth noting that Grant and Kim (2007) do not state which, if any, of these individual treatments involved adolescents.

Given the lack of empirically supported psychotherapeutic interventions for individuals with pyromania, most clinicians will draw on broader treatment options available for firesetting behavior more generally. There are only two standardized treatment programs currently available for adult firesetters in the UK (Gannon et al., 2015; Gannon, Lockerbie, & Tyler, 2012) and none available in the USA (Gannon & Pina, 2010). There are, however, many more programs and initiatives available for use with children and adolescents. We do not examine these in detail here since they have not been developed specifically to address pyromania (see instead: MacKay et al., 2012; Kolko, 2002). To give one example, however, Kolko (2001) examined the efficacy of three different approaches with a sample of children who had set a recent fire. Cognitive-behavioral treatment (CBT) was compared with fire safety education (FSE) and a home visit from a firefighter (HVF). Results showed that children benefited from all three interventions but that the strongest gains were made for those who took part in CBT or FSE. Gains included reductions in frequency of firesetting, improved regulation of inappropriate fire use, and a reduction in the proportion of children showing fire interest.

Prognosis and Outcome

The available evidence makes it extremely difficult to provide an accurate prognosis for young people with treated or untreated pyromania. While various psychotherapeutic interventions have shown efficacy in reducing firesetting behavior generally, there has been no systematic evaluation of their efficacy with children or adults with pyromania. Therefore, while these interventions may have had group-level positive effects for groups which included individuals with pyromania, it remains unclear whether young people diagnosed with pyromania would have shown the same level of improvements. As mentioned previously, while five individuals in Grant and Kim's (2007) study no longer met the criteria for pyromania, suggesting that pyromania symptoms do dissipate, each went on to develop symptoms of another impulsive or compulsive disorder. Additionally, they reported that for the majority of individuals, frequency and severity of firesetting increased over the course of the illness. Both these findings are suggestive of typically poor outcomes for adults with pyromania. Based on the current state of the research, we can say very little about the outcomes for children with pyromania. Clearly, further systematic research is required before an accurate conclusion can be drawn regarding prognosis for pyromania.

Conclusion

MacKay et al. (2012) highlight that deferral "to antisociality or conduct problems as the primary 'diagnostic ballpark' for youth with firesetting may have inadvertently limited research on the role of fire-specific pathology in firesetting" (p.848). They go on to state that while conduct disorder may be the most frequent diagnosis given to juvenile firesetters, it is neither necessary nor sufficient to explain firesetting behavior. It remains unclear, however, whether pyromania is a useful alternative diagnosis to explain pathological firesetting in children and

adolescents. Research on adult pyromania is plagued by uncontrolled studies using convenience samples. However, research on pyromania in children and adolescents is almost nonexistent. As mentioned, this may be due to the true base rate of the disorder in younger individuals, or it may be due to reluctance to stigmatize young people with a label of pyromania. Alternatively, the sheer number of exclusionary criteria in successive editions of the DSM may have rendered the consulting clinician's task so difficult that diagnoses such as conduct disorder may offer a simpler explanation of firesetting behavior, even where the essential characteristics of pyromania are present.

We suggest that a change is required in research on pyromania in order to provide a robust empirical basis for its retention in future editions of the DSM. In particular, prospective cohort studies are required to determine true prevalence rates and to examine the etiology, onset, and developmental course of the disorder. Furthermore systematic research must be carried out examining the validity of each of the exclusionary criteria stated in DSM-5. The somewhat simplistic assumption that firesetters may not have pyromania if another factor operated as an additional motivating factor (e.g., criminal activity) is likely to result in the underdiagnosing of the disorder. Without substantial research on pyromania in young people and into adulthood, pyromania within the DSM remains little more than a supplementary term despite numerous editions and text revisions and despite the enormous human and financial cost of deliberate firesetting.

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28 Alcohol-Related Disorders and Other Substance Use Disorders

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Overview of the History and Current Diagnostic Presentation

Substance use disorders have undergone some changes from DSM-IV to DSM-V. DSM-V has combined the previous categories of substance abuse and substance dependence into a single disorder (substance use disorder). Furthermore, drug craving has been added to the criteria, while problems with law enforcement have been removed due to cultural biases. The use disorders specify whether the disorder is mild, moderate, or severe. For each specific substance (other than caffeine), there is a separate category (e.g., alcohol use disorder, cannabis use disorder) with similar diagnostic criteria. Furthermore, whereas the diagnosis of substance abuse only required one symptom in previous criteria, individuals are now required to meet at least two out of 11 symptoms in order to meet the specification of mild. In DSM-V moderate severity is indicated with meeting four to five symptoms, and severe severity is diagnosed when six or more symptoms are met (Agrawal et al., 2014; American Psychiatric Association [APA], 2013; Fitzgerald & Morgan, 2012; Hasin et al., 2013).

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All of the substances have diagnostic criteria pertaining to impaired control, social impairment, risky use, and pharmacological criteria. Impaired control covers Criteria 1–4 and consists of taking a substance in larger amounts or over a longer period than was originally intended (Criterion 1) and persistent desire to cut down or regulate substance use that may report multiple unsuccessful efforts to decrease or discontinue use (Criterion 2) spending a great deal of time obtaining the substance; using the substance, or recovering from its effects (Criterion 3); and craving as manifested by an intense desire or urge for the drug that may occur at any time but is more likely when in an environment where the drug previously was obtained or used (Criterion 4). Social impairment covers Criteria 5–7. These criteria consist of recurrent substance use that may result in a failure to fulfill major role obligations at work, school, or home (Criterion 5); the individual may continue use despite having persistent, recurrent social, or interpersonal problems caused or exacerbated by the effects of the substance (Criterion 6); and important social, occupational, or recreational activities may be given up or reduced because of substance use (Criterion 7). Risky use of the substance consists of Criteria 8–9. The criterion consists of recurrent substance use in situation in which it is physically hazardous (Criterion 8), and the individual may continue substance 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

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(Criterion 9). Finally, the pharmacological criteria consist of Criteria 10–11. Criterion 10 consists of tolerance, while Criterion 11 consists of withdrawal. The criteria are not organized hierarchically, and the criteria all carry equal weight when diagnosing. Further, depending on the substance, there are subcategories concerned with use, intoxication, and withdrawal (APA, 2013; Kelly et al., 2014; Parritz & Troy, 2014).

Alcohol-Related Disorders

Alcohol use disorder as it is seen in the DSM-V, Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (APA, 2013), is not the same as it has been portrayed in previous additions of the DSM. For example, prior to the publication of DSM-III in 1980, this disorder was categorized as alcoholism under personality disorders. In the second edition of the DSM, alcoholism became its own disorder though still grouped with personality disorders. It was also in this edition that there were three subtypes of alcoholism consisting of episodic excessive drinking, habitual excessive drinking, and alcoholic addiction (Durrant & Thakker, 2003). Furthermore, the first and second editions of DSM did not present explicit diagnostic criteria for alcoholism or any other disorder for that matter. In its place, each disorder was explained in a brief paragraph, which covered a broad description of the disorder. However, the DSM-III did include specific diagnostic criteria for the first time. It also included two alcohol use disorders instead of alcoholism, consisting of abuse and dependence. Changes from DSM-III to DSM-IV on alcohol-related disorders were minor. The only alterations consisted of some constraints on the DSM-IV dependence category and an expansion of DSM-IV's abuse category criteria (Hasin, 2003). There have been notable changes from DSM-IV to DSM-V in alcohol-related disorders. For example, DSM-V combines the DSM-IV categories of substance abuse and substance dependence into a single disorder called alcohol use disorder (AUD), measured on a continuum from mild to severe (APA, 2013; National Institute on Alcohol Abuse and Alcoholism [NIAAA], 2013).

Current Diagnostic Criteria of Alcohol Use Disorder Alcohol use disorder is diagnosed in individuals who have a pattern of alcohol use that meets Criterion A. Criterion A is met when there is a problematic pattern of use that causes distress or substantial impairment as manifested by at least two of eleven symptoms. Anyone meeting at least two of eleven criteria during a 12-month period would receive a diagnosis of AUD. Criteria A.1–A.11 are as follows:

1. Alcohol is taken in larger amounts or over a longer period than was intended.
2. Persistent desire or unsuccessful efforts to cut down or control use.
3. A great deal of time is spent in activities necessary to obtain, use, or recover from alcohol.
4. Craving or strong desire to use.
5. Recurrent alcohol use resulting in a failure to fulfill major role obligations at work, school, or home.
6. Continued use despite having persistent or recurrent social or interpersonal problems caused or exacerbated by the effects of alcohol.
7. Important social, occupational, or recreational activities are given up or reduced due to alcohol use.
8. Recurrent use in situations in which it is psychically hazardous.
9. Use is continued despite knowledge of having a persistent or recurrent physical or psychological problem that is likely to have been caused or exacerbated by alcohol.
10. Tolerance, as defined by (a) a need for increased amounts of alcohol to achieve intoxication or desired effect or (b) a markedly diminished effect with continued use of the same amount of alcohol.
11. Withdrawal, as manifested by either (a) the characteristic withdrawal syndrome for alcohol or (b) alcohol (or related closely substance) that is taken to relieve or avoid withdrawal symptoms (APA, 2013; NIAAA, 2013).

Current Diagnostic Criteria of Alcohol Intoxication Alcohol intoxication is diagnosed when four criteria are met. Criterion A is that there was a recent ingestion of alcohol. Criterion B pertains to clinically significant problematic behavioral or psychological changes (e.g., inappropriate sexual or aggressive behavior, mood lability, impaired judgment) that occur during or shortly after the ingestion of alcohol. Criterion C is met when one or more of the following signs or symptoms develop during or shortly after alcohol use: slurred speech, incoordination, unsteady gait, nystagmus, impairment in attention or memory, or stupor or coma. Finally, Criterion D consists of the indication that the signs or symptoms are not caused by another medical condition or are not better explained by another mental disorder, including intoxication with another substance (APA, 2013).

Current Diagnostic Criteria of Alcohol Withdrawal Alcohol withdrawal is diagnosed when four criteria are met. Criterion A is that the cessation or reduction in alcohol use after heavy and prolonged use has occurred. Criterion B is met when two or more symptoms (i.e., autonomic hyperactivity; sweating or pulse rate greater than 100 bpm; increased hand tremor; insomnia; nausea or vomiting; transient visual, tactile, or auditory hallucination or illusions; psychomotor agitation; anxiety; or generalized tonic-clonic seizures) develop within several hours to a few days after cessation or reduction in alcohol. Criterion C is that the symptoms must cause clinically significant distress or impairment in social and occupational functioning or other important areas of functioning. Finally, Criterion D is that the signs or symptoms must not be attributable to another medical condition and are not better explained by a different mental disorder, including intoxication or withdrawal from another substance (APA, 2013).

Tobacco-Related Disorder

Tobacco-related disorders have undergone various modifications in the DSM-V. It was decided that tobacco use disorder's criteria should be aligned with the criteria of the other substance use disorders. Therefore, it changed from being nicotine dependence in DSM-IV to tobacco use disorder (Hasin et al., 2013). Prior to DSM-IV there was a classification for nicotine abuse and dependence. In DSM-IV nicotine abuse was taken out because experts decided the abuse criteria were not applicable to nicotine (Hasin et al., 2013; Martin, Chung, & Langenbucher, 2008).

Current Diagnostic Criteria of Tobacco Use Disorder This disorder is diagnosed when there is a problematic pattern of tobacco use leading to clinically significant impairment or distress. Criteria A is met when at least two of 11 specified symptoms occur within a 12-month period. Criteria 1–11 are the same as with alcohol use disorder (APA, 2013).

Cannabis-Related Disorder

There have been minimal changes in cannabis-related disorders over time. Cannabis withdrawal was not incorporated in DSM-IV due to lack of evidence to support its inclusion. Recently, however, reliability and validity of cannabis withdrawal has been established in preclinical, clinical, and epidemiological studies. Therefore, in DSM-V cannabis withdrawal disorder was added, and withdrawal was also included as a criterion for cannabis use disorder (Hasin et al., 2013; Martin et al., 2008).

Current Diagnostic Criteria of Cannabis Use Disorder This disorder is diagnosed when there is a problematic pattern of cannabis use leading to clinically significant impairment or distress. Criterion A is met when at least two of 11 specified symptoms occur within a 12-month period. Criteria 1–11 are the same as with alcohol use disorder (APA, 2013).

Current Diagnostic Criteria of Cannabis Intoxication This disorder is diagnosed when four criteria are met. Criterion A is that the individual has recently used cannabis. Criterion

B is met when there are clinically significant problematic behavioral or psychological changes (e.g., impaired motor coordination, euphoria, anxiety, sensation of slowed time, impaired judgment, social withdrawal) that occur during or shortly after the use of cannabis. Criterion C is met when two or more of the following symptoms occur within 2 hours of cannabis use: conjunctival injections, increased appetite, dry mouth, and tachycardia. Criterion D states that the signs or symptoms are not attributable to another medical condition and are not better explained by another mental disorder. Clinician should specify if the disorder occurs with perceptual disturbances, such as hallucinations (APA, 2013).

Current Diagnostic Criteria of Cannabis Withdrawal The diagnostic criteria for cannabis withdrawal consist of four criteria that must be met. Criterion A is that cessation of cannabis use that has been heavy and prolonged (i.e., usually daily or almost daily use over a period of at least a few months). Criterion B is met when three or more of the following symptoms develop 1 week after Criterion A: irritability, anger or aggression, nervousness or anxiety, sleep difficulty, decreased appetite or weight loss, restlessness, depressed mood, and at least one of the following physical symptoms (abdominal pain, shakiness/tremors, sweating, fever, chills, or headache). Criterion C is that the symptoms in Criterion B cause clinically significant distress or impairment in social and occupational functioning or other important areas of functioning. Finally, Criterion D is met if the symptoms are not attributable to another medical condition or mental disorder, including intoxication or withdrawal from another substance (APA, 2013).

Other Substance Use Disorders

Phencyclidine, inhalants, opioids, sedative, hypnotic, anxiolytic, and stimulants will not appear differently in DSM-V than in DSM-IV. The substances have changed from being separated into dependence and abuse (e.g., stimulant dependence and stimulant abuse) into a single use disorder (e.g., stimulant use disorder) (APA, 2013). This separation of abuse and dependence first appeared in DSM-III (Jones, Gill, & Ray, 2012). It is also in the third edition where substance use disorders got their own section for the first time. Before this edition, drug addiction was grouped with "sociopathic personality disturbances" (APA, 1952, 1980).

Current Diagnostic Criteria of Phencyclidine Use Disorder

Diagnosis of this disorder occurs when an individual uses phencyclidines (or phencyclidine-like substances) including phencyclidine (e.g., PCP, "angel dust") and other similarly acting compounds such as ketamine. These compounds are known as hallucinogens due to their mind-altering effects. Individuals are diagnosed if they exhibit a pattern of use leading to significant impairment manifested by at least two of ten symptoms within a 12-month period. Diagnostic criteria are the same as alcohol use disorder except it does not include withdrawal. Clinicians should also specify if this disorder is in early remission or in sustained remission and specify if the individual is in a controlled environment where access to phencyclidine use is restricted. Clinicians should also specify the current severity (APA, 2013).

Current Diagnostic Criteria of Inhalant Use Disorder Inhalant use disorder is diagnosed when an individual uses hydrocarbon-based inhalant substances. Diagnostic criteria are the same as with phencyclidine use disorder. Clinicians are to specify the particular inhalant involved (e.g., "solvent use disorder"). They should also specify if this disorder is in early remission or in sustained remission and specify if the individual is in a controlled environment where access to inhalant substances is restricted. Clinicians should also specify the current severity (APA, 2013).

Current Diagnostic Criteria of Opioid Use Disorder Opioid use disorder is diagnosed when there is a prolonged self-administration of opioid substances without a legitimate medical purpose or the dosage greatly exceeds the needed amount. Diagnosis is based on whether the individual meets at least two of 11 symptoms within a 12-month period. Criteria 1–11 are the same as with alcohol use disorder. Clinicians should specify if the individual is in early remission or in sustained remission, if they are on maintenance therapy or in a controlled environment. They should also specify current severity (APA, 2013).

Current Diagnostic Criteria of Sedative-, Hypnotic-, or Anxiolytic-Related Disorders Sedative, hypnotic, or anxiolytic substances include benzodiazepines, benzodiazepine-like drugs, carbamates, and barbiturate-like hypnotics. This class of substances includes all prescription sleeping medication and most prescription antianxiety medication. Diagnosis is based on whether the individual meets at least two of 11 symptoms within a 12-month period. Criteria 1–11 are the same as with alcohol use disorder. Clinicians should specify if the individual is in early remission or in sustained remission, if they are on maintenance therapy or in a controlled environment. They should also specify current severity (APA, 2013).

Current Diagnostic Criteria of Stimulant Use Disorder Stimulant use disorder can be diagnosed with the use of amphetamine and amphetamine-type stimulants such as amphetamine and methamphetamine. This diagnosis also includes substances that have different chemical structures such as cocaine. An individual meets the diagnostic criteria when they exhibit a pattern of stimulant use leading to clinically significant impairment. They must show at least two of 11 symptoms within a 12-month period. Criteria 1–11 are the same as with alcohol use disorder. Clinicians should specify if patients are in early remission or in sustained remission and specify severity if in a controlled environment. They should also specify the substance type of amphetamine, cocaine, or other unspecified stimulant (APA, 2013).

Research Review

Etiology of General Substance Abuse

Substance abuse is the result of a complex interplay of multiple genetic, person-level, contextual factors (Zucker, 2006). The following review of the etiology of substance use disorders will review the literature related to genetic and person-level risk factors, contextual risk factors, and their interrelationships and contribution to the development of substance use disorders.

Genetic and Person-Level Risk Factors While genetics plays a role in the development of substance use disorders (SUDs), there is no single gene that has been pinpointed to contribute to its development. Rather, a multitude of genes with individual effects interact with each other as well as with an individuals' environment to make one more or less susceptible to the development of SUDs (Meyers & Dick, 2010). Genetic influences can determine an individual's personality traits and further risk for substance abuse (Hopfer, Crowley, & Hewitt, 2003). For example, personality traits such as negative emotionality and behavior disinhibition have been found to be risk factors for later substance abuse (Hicks et al., 2013; Feske et al., 2008). Additionally, a pattern of antisocial behavior during childhood is a consistent and strong predictor of substance abuse (Iacono, Carlson, Taylor, Elkins, & McGue, 1999; Zucker & Gomberg, 1986). The combination of antisocial behavior and personality traits such as behavioral disinhibition is often referred to as the behavioral disinhibition pathway to substance abuse. This pathway is associated with earlier-onset and more severe and persistent substance abuse (Hicks et al., 2013).

Other related person-level factors found to be predictive of adolescent substance abuse include low levels of socialization and boldness (Hicks et al., 2013). Low levels of socialization refers to

an individual's willingness to follow rules and endorse conventional moral and ethical values, while boldness refers to a person's propensity to seek out thrilling situations with a lack of anxiety and fear. Each has a unique pathway to adolescent substance abuse with boldness exhibiting high levels of genetic influence and no environmental influences and low socialization having both moderate heritability and environmental influences (Hicks et al., 2013). Both of these factors taken together present a comprehensive picture of the risk for adolescent substance abuse (Hicks, Iacono, & McGue 2014).

Contextual Risk Factors The development of substance use disorders during adolescence is also highly influenced by a number of contextual risk factors including family, peer, school, and neighborhoods (Hicks et al., 2013). These risk factors contribute to early substance use initiation and subsequent adolescent substance abuse. Hicks et al. (2013) discuss the interaction of these risk factors and their contribution to adolescent substance abuse as a "developmental cascade." Contextual risk factors for adolescent substance abuse include parental conflict, low parental monitoring, neighborhoods with high rates of crime, poverty, family transience, and residential instability (Meyers & Dick, 2010; Hicks et al., 2013). Due to the nature of these risk factors, individuals may be exposed to multiple risk factors throughout their childhood. Exposure to these risks varies by age group with exposure peaking from ages 11 to 14 and with boys experiencing greater exposure than girls (Hicks et al., 2013).

Interaction Between Child Level and Contextual Factors Individual person-level risk factors also determine one's potential exposure to contextual risk factors. Hicks et al. (2014) found that low socialization at age 11 was highly correlated with concurrent contextual risk factors including antisocial peer affiliation, academic failure and disengagement, lack of prosocial peers, poor parent-child relationships, and stressful life events impacting family context (Hicks et al., 2013). Therefore, person-level risk factors put one at risk for additional contextual risk factors, and these relationships ultimately manifest in substance use and abuse in adolescence (Hicks et al., 2013).

Environmental factors have also been shown to moderate the influence of genetic factors on substance abuse. Some moderating variables include childhood stressors, access to drugs, antisocial and prosocial peer groups, religiosity, parental attitudes toward drugs, parental monitoring, and socioregional factors (Meyers & Dick, 2010). Meyers and Dick (2010) investigated the influence of these moderating factors and found that religiosity moderates the relationship between genetics and alcohol use among females and that genetic factors have a larger impact on substance use behaviors for those without religious upbringing. Little parental monitoring, large numbers of peers who use substances, and neighborhoods with less stability all contributed to higher levels of genetic influence and increased substance use (Meyers & Dick, 2010). He concludes that the presence of contextual risk factors increases the importance of genetic influences on the etiology of externalizing disorders and substance abuse (Meyers & Dick, 2010).

Associated Impairments and Comorbidities

It has been estimated that approximately 75% of adolescents with substance use disorders also have coexisting disorders with externalizing disorders such as conduct disorder and ADHD followed by affective disorders (Crowley, Macdonald, Whitmore, & Mikulich, 1998; Greenbaum, Foster-Johnson, & Petrila, 1996). These disorders are both predictors and consequences of substance use (Costello, 2007). Youth with comorbid disorders often experience more severe substance use behaviors, use a greater number of substances, and initiate drug use at earlier ages (Grella, Hser, Joshi, & Rounds-Bryant, 2001).

Externalizing

Conduct Disorder

The comorbidity between substance use disorders and conduct disorder has a robust literature base. Meyers and Dick (2010) found that the majority of the genetic variance on alcohol and illicit drug dependence resulted from a shared genetic factor with externalizing psychopathology (Meyers & Dick, 2010). Others suggest that early symptoms of conduct disorder are an expression of a genetic trait that is also a risk factor for later substance use disorders (Tarter et al., 1999). Longitudinal studies support these assertions finding that early conduct disorder symptoms are associated with earlier-onset and more frequent and intense substance use across time (Weinberg, Rahdert, Colliver, & Glantz, 1998). Thus, early detection of conduct disorders may identify those adolescents who are at highest risk for later substance problems (Flory Milich, Lynam, Leukefeld, & Clayton, 2003; Miller-Johnson, Lockman, Coie, Terry, Hyman, 1998).

Attention Deficit Hyperactivity Disorder Approximately a quarter of individuals with substance use disorders meet criteria for ADHD (van Emmerik-van Oortmerssen et al., 2012). Childhood ADHD is a major risk factor for the development of substance use disorders, either directly or through its relationship with conduct disorder (van Emmerik-van Oortmerssen et al., 2012). Studies find that children with ADHD were more likely to develop substance use disorders with nicotine, alcohol, marijuana, cocaine, and other substances (Lee, Humphreys, Flory, Liu, & Glass, 2011). The gateway hypothesis asserts that children with ADHD are more likely to use nicotine and that becomes a gateway to more illicit drugs (Kandel, 2002). While some find a direct relationship between ADHD and substance use disorders (Wilens et al., 2011), others state that this relationship dissipates when you consider the effect of other externalizing behaviors such as conduct disorder (Looby, 2008; Lee et al., 2011).

Internalizing

The majority of research looking at comorbidity between mental health disorders and substance use disorders focuses on externalizing disorders. While strong relationships exist between externalizing problems and substance use, internalizing disorders should also be considered when looking at substance use. Internalizing disorders such as anxiety and depressive are among the most common disorders of childhood and adolescence and are often comorbid with substance use disorders in adolescence (Marmorstein, 2012; O'Neil, Connor, & Kendall, 2011). Depression is the second most commonly comorbid disorder with substance use disorders with rates ranging from 11% to 32% (Armstrong & Costello, 2002; O'Neil et al., 2011). The rate of comorbidity between anxiety and substance use disorders ranges from 7% to 40% (O'Neil et al., 2011). Overall rates of comorbidity between internalizing disorders and SUDs range from 11% to 48% with higher rates for depressive disorders than anxiety disorders (O'Neil et al., 2011).

The majority of research in this area supports a unidirectional model in which childhood internalizing disorders increase risk for later substance use disorders (O'Neil et al., 2011), while other research finds the relationship between internalizing disorders and later substance abuse disappears when you control for prior adolescent substance use (Brook, Cohen, & Brook, 1998; Rohde, Lewinsohn, & Seeley, 1991). In clinical adolescent samples, about half had a current comorbid or anxiety disorder (Deykin, Buka, & Zeena 1992; Lubman, Allen, Rogers, Cementon, & Bonomo, 2007). Sex differences have also been found for the relationship between SUDs and internalizing disorders. Female adolescents are at a greater risk for comorbidity between internalizing and substance use disorders than males (Clark et al., 1997; Deykin et al., 1992).

Other Substances

Many individuals who have a substance use disorder use multiple substances, with high rates of comorbidity across various drug classes (Meyers

& Dick, 2010). Using one substance increases the odds of using any other substance by at least 50 times (Roberts, Roberts, Xing, 2007). Common genetic factors have been found to be shared across alcohol dependence and illicit drug dependence as well as childhood externalizing disorders (Kendler, Prescott, Myers, & Neale, 2003).

Course and Developmental Challenges

Risk factors for the development of adolescent and adult substance use disorders are often present in childhood and contribute to a trajectory of risk-related behavior culminating in the initiation and subsequent abuse of substances (Meyers & Dick, 2010). The age of initiation of substances contributes significantly to the course and development of substance use disorders with those using earlier having a higher likelihood of developing SUDs. The prevalence of substance use and abuse increases dramatically with age, with those 12 and younger having the lowest rates, followed by those 13 to 15 with intermediate rates and those 16 and older having the highest rates of substance abuse (Roberts et al., 2007). By twelfth grade, high school students report the highest rates of using alcohol (51%), followed by tobacco (34%), marijuana (22%), hallucinogens (3.5%), and cocaine (2%) (Johnston, O'Malley, & Bachman, 2001). About 7% of adolescents aged 12–17 meet diagnostic criteria for substance use disorders (US Department of Health and Human Services, 2011), and approximately 145,000 adolescents enroll annually in outpatient substance use treatment nationwide (Substance abuse and mental health services administration [SAMHSA], 2007). Adolescent substance use is a risk factor of adult substance use and antisocial personality disorder (Meyers & Dick, 2010).

Comorbidity rates also vary by age with those 13–15 having significantly higher rates of comorbid internalizing and externalizing behavior disorders (Roberts et al., 2007). Comorbidity has important implications for the course and development of substance use disorders. For example, individuals with both ADHD and SUDs become addicted at younger ages, use more substances, and are hospitalized more often than SUD patients without ADHD (Arias et al., 2008).

Treatment

There are several evidence-based approaches to the treatment of adolescent substance use including family therapies, cognitive behavioral therapies, and newer approaches such as motivational interviewing.

Family Therapy Family therapy is touted as a well-established evidence-based treatment for adolescent substance abuse. There are various types of family therapy with varying levels of evidence base. Functional family therapy and multidimensional family therapy are well-established treatment approaches, while behavioral family therapy, brief strategic family therapy, and multisystemic therapy are probably efficacious and in need of additional study (Waldron & Turner, 2008). Family-based treatment is the most common treatment provided to adolescents with substance use disorders and contributes to significant reductions in substance use among adolescents (Liddle & Dakof, 1995; Stanton & Shadish, 1997; Waldron, 1997). Family-based treatment has been shown to have a greater impact than group therapy, family education, individual therapy, tracking, or juvenile justice system interventions on reducing adolescent substance use (Waldron, Slesnick, Brody, Turner, & Peterson, 2001). A combination of both family and individual therapy results in reductions in alcohol use as well (Waldron et al., 2001). Cited limitations of family-based treatment include a lack of attention placed on intrapersonal risk factors related to substance abuse including coping strategies, coping with urges, and peer refusal (Waldron et al., 2001).

Cognitive Behavioral Therapy Cognitive behavioral therapy can be provided in two different formats: group and individual. Both cognitive behavioral therapy–individual (CBT-I) and cognitive behavioral therapy–group (CBT-G) are well-established evidence-based treatments

(Hogue, Henderson, Ozechowski, & Robbins, 2014). Cognitive behavioral therapy addresses both individual and intrapersonal factors related to substance abuse. Adolescents identify behavioral goals and develop self-regulation and coping behaviors. Interventions may include ways to cope with urges, how to refuse peer advances, and problem-solving skills. While both CBT-I and CBT-G are both evidence based, individual therapy is more individualized and may produce more significant treatment outcomes (Waldron et al., 2001). Limitations of cognitive behavioral approaches are that they neglect the family component and the multitude of risk factors that stem from family and ecological contexts.

Other Approaches Another promising approach is motivational interviewing (Macgowan & Engle, 2010; Barnett, Sussman, Smith, Rohrbach, Spruijt-Metz, 2012; Hogue et al., 2014). Motivational interviewing is a client-centered counseling style that explores and resolves ambivalence about changing personal behaviors (Miller & Rollnick, 2002). Motivational interviewing is an ideal therapy technique to use with adolescents asserting their independence as it assists the adolescent with their decision-making and intrinsic motivation toward healthy behaviors (Baer & Peterson, 2002; Naar-King & Suarez, 2011). Additional approaches such as drug counseling traditional educational approaches or approaches combining a number of strategies are thought to be promising treatment approaches but in need of additional research support (Waldron et al., 2001; Hogue et al., 2014).

Treatment Considerations New advances in research are finding new and innovative ways to treat substance abuse. Some new treatment approaches focus on prevention and treating earlier risk factors for adolescent substance abuse such as externalizing or internalizing disorders. Initial studies in this area provide support for treating internalizing or externalizing disorders as a way to prevent further development of substance use disorders (Puleo, Conner, Benjamin, & Kendall, 2011). Additional studies are looking at gene-specific (i.e., traits of impulsivity) interventions to tailor interventions to specific personality traits to improve substance use disorder outcomes (Meyers & Dick, 2010). Lastly, it is important to consider the population you are working with when implementing interventions as research has found that treatments may be less efficacious for Hispanic youth (Waldron & Turner, 2008).

Prognosis and Outcome

Epidemiological studies report that the lifetime prevalence for alcohol use disorders is 13.5% while the prevalence of other drug use disorders is 6.1% (van Emmerik-van Oortmerssen et al., 2012). Substance use disorders during adolescence have particularly damaging consequences such as the increase of motor vehicle accidents and other injuries (Lynskey, Agrawal, & Heath, 2010). The prognosis and outcomes for adolescents who seek treatment for substance use disorders are promising. While promising, there are a number of hurdles adolescents must overcome to live successful adult lives. Substance abuse during adolescence is associated with a more severe and persistent course of abuse and also increases the risk of adult antisocial behavior (Clark, Kirisci, Tarter, 1998; Hicks, Iacono, McGue, 2010). Additionally, the severity of drug use is associated with earlier treatment initiation, repeated treatment utilization, and poorer treatment outcomes (Anglin, Hser, & Grella, 1997; Hser, Joshi, Anglin, & Fletcher, 1999).

The research on the impact of comorbidity on treatment outcomes is mixed. Some researchers find that adolescents who have additional comorbid disorders may have difficulty recovering from substance abuse and experience more severe substance use prior to treatment and higher relapse rates after treatment (Brown, Myers, Mott, & Vik, 1994; Carroll and Rounsaville, 1993; Kennedy & Minami, 1993). Other research on comorbidity between internalizing and substance use disorders find that those with both depression and substance use disorders have comparable

treatment outcomes to those with just depression only (Rohde, Clarke, Lewinsohn, Seeley, & Kaufman, 2001; Rohde, Seeley, Kaufman, Clarke, & Stice, 2006).

Alcohol Use Disorders

Etiology

The etiology of alcohol use disorders shares many similarities to the etiology or development of all substance use disorders. Alcoholism is influenced by both genetics and environmental factors such as familial environments, schools, peers, and neighborhoods (Meyers & Dick, 2010). Research finds that the various risk factors for the development of alcohol use disorders vary in their contribution to the development of alcohol use disorders over time. The etiologic model, for example, asserts that early alcohol use is more strongly influenced by ecological factors while later use or initiation is more strongly influenced by genetics (Kendler, Schmitt, Aggen, & Prescott, 2008). It is hypothesized that genetics are not able to be expressed during early adolescence when the majority of activities are governed by parents. Thus, early decisions to drink are thought to be more an outcome related to environmental factors. However, later alcohol initiation and use (through emerging adulthood) are thought to be expressions of genetic predispositions due to the decrease of parental influence and the increase of independent decision-making (Meyers & Dick, 2010).

Associated Impairments and Comorbidities

A number of studies have connected the presence of alcohol use disorders and other mental health disorders. The following will review the literature on associated externalizing and internalizing mental health disorders. First is a review of externalizing mental health disorders such as conduct disorder and attention deficit hyperactivity disorder. This will be followed by a review of internalizing disorders that include anxiety and depression.

Externalizing Disorders

The relationship between externalizing disorders and alcohol use disorders is well established. Researchers find that for those with disruptive disorders, males are 9.3 times more likely to develop alcohol dependence, while females are 19.2 times more likely to develop alcohol dependence if disruptive behaviors are present (Roberts et al., 2007). The relationship between externalizing behaviors and alcohol use is so inextricably linked, and some researchers have asserted that early use of alcohol may be an indicator of overall risk and may disrupt adolescent development (McGue & Iacono, 2008). Further, research has found that alcohol use prior to age 15 is associated with a range of externalizing behavior problems including conduct disorder and attention deficit hyperactivity disorder (Meyers & Dick, 2010).

Conduct Disorder Conduct disorder is perhaps the most closely linked mental health disorder to alcohol use. Research overwhelmingly indicates that both alcohol use disorders and conduct disorder are strongly linked (Weinberg et al., 1998). In addition, researchers find that the risk and correlation between these two mental health disorders is reciprocal. For example, some researchers find that adolescent substance use is a risk factor for later development of conduct problems in adolescence (Wymbs et al., 2014), while other research finds that early conduct disorder symptoms predict later alcohol use disorder symptoms and diagnoses by young adulthood (Pardini, White, Stouthamer-Loeber 2007). As the result of this link, researchers have hypothesized pathways of risk between childhood conduct disorder and later alcohol problems. They hypothesize that the relationship between these two mental health disorders is a result of shared genetic factors. Similar genes have been found in both early conduct disorder and separate studies investigating adolescent or adult alcohol dependence. Thus, early conduct problems may express an underlying disposition to later alcohol problems (Meyers & Dick, 2010). Longitudinal research supports these hypotheses finding that early conduct dis-

order symptoms are associated with more frequent and intense alcohol use over time (Weinberg et al., 1998).

Attention Deficit Hyperactivity Disorder The research on the relationship between attention deficit hyperactivity disorder (ADHD) and alcohol use disorders is mixed. Some researchers find that nearly a quarter of adults aged 18–44 with substance use disorders have comorbid ADHD (Charach, Yeung, Climans, & Lillie, 2011). They also find that childhood ADHD significantly increases the risk for alcohol use during young adulthood (Charach et al., 2011). Other researchers conclude, however, that the relationship between ADHD and alcohol use disorders disappears when you control for other co-occurring conduct disorders (Pardini et al., 2007).

Internalizing Disorders

While externalizing disorders have proven to have a strong relationship with alcohol use disorders, internalizing disorders such as anxiety and depression have also been found to be comorbid with alcohol use disorders. Roberts et al. (2007) found that the likelihood of the presence of a mood disorder for individuals with alcohol dependence is 80% for females and 3.7% for males. In addition, Stice, Barrera, and Chassin (1998) found that internalizing symptoms were predictive of problem alcohol use for adolescents even after controlling for consumption levels.

Anxiety Research on anxiety and alcohol use disorders has found some comorbidity between these two mental health disorders. The odds of anxiety disorders has been found to increase for those with alcohol dependence, and some studies cite the lifetime comorbidity between anxiety and alcohol dependence to be 17% (Roberts et al., 2007; Rohde, Lewinsohn, & Seeley, 1996). Other studies find no relationship between anxiety symptomatology and alcohol use (Pardini et al., 2007; O'Neil et al., 2011). O'Neil posits that this could be due to the heterogeneity of anxiety disorders offsetting the effects of specific anxiety disorders. Thus, while specific anxiety disorders such as generalized anxiety disorder predict higher risk of alcohol use in adolescence, separation anxiety symptoms predict lower risk of alcohol use (O'Neil et al., 2011). Therefore, looking at specific anxiety disorders is key when looking at comorbidity.

Depression Comorbidity between depression and problematic alcohol use is around 50% (Rohde et al., 1996). Studies find a strong relationship between depression and alcohol use disorders. This is true for both children (O'Neil et al., 2011) and adolescents (White, Xie, Thompson, Loeber, & Stouthamer-Loeber, 2001), with research finding early symptoms of depression predicting later alcohol use. This relationship remains even after controlling for other mental health issues such as conduct problems (Henry et al., 1993; Sung, Erkanli, Angold, & Costello, 2004). However, having both depression and conduct disorder symptoms places adolescent boys at significant risk for developing later alcohol use disorder symptoms and diagnoses (Pardini et al., 2007).

Course and Developmental Challenges

The prevalence of alcohol use disorder is high in the United States. In adolescence (ages 12–17), the estimated occurrence is 4.6% (APA, 2013; Parritz & Troy, 2014). The trend in alcohol use involves a gradual increase during adolescence, a peak in early adulthood, and then a steady decrease. The age of first use is a critical factor. Those who start drinking early are at an increased risk of later substance abuse (13.5%) and dependence (15.9%) (DeWit, Adlaf, Offord, & Ogborne, 2000). Furthermore, early alcohol consumption is associated with increased rates of binge drinking, risky sexual activity, aggression and violence, and unfavorable adult outcomes (Parritz & Troy, 2014). Alcohol intoxication occurs frequently in the adolescent population. For example, in 2010, 44% of twelfth grade students stated they had been drunk in the past year. The first incidence of alcohol intoxication tends to occur around the age of 15 (Wittchen et al., 2007). By the age of 18, 60% of

teenagers have experienced drunkenness (APA, 2013; Schuckit, 2009). The earlier the onset of regular alcohol intoxication, the greater the likelihood that the individual will later meet the diagnostic criteria for alcohol use disorder (Grant & Dawson, 1998). Unfortunately, the age of onset for alcohol use disorder peaks in late teens and mid-twenties.

There are three theories related to the course and development of alcohol use disorders during childhood and adolescence. The first theory argues that early alcohol exposure and use could expose youth to deviant peers who then reinforce future deviant behavior. The second theory posits that early use of alcohol changes biological development through the changing of the adolescent brain. The last theory connects the association of early alcohol use and later alcohol use disorders via a shared general genetic lack of inhibitory controls (Meyers & Dick, 2010).

Treatment

In the past, treatment for adolescents with alcohol disorders tended to consist of the same treatments for adults. However, recent treatment has improved by taking into account development in order to specifically target adolescents. For example, it is believed that developmental differences between adults and adolescence can impact treatment, adherence, and outcomes (Deas, 2008). Treatments that have been found to be effective for the adolescent population consist of family-based interventions, motivational enhancement therapy, behavioral therapy, and cognitive behavioral therapy (Deas, 2008). In addition, treatment of co-occurring disorders such as ADHD has been associated with lower rates of substance use by age 18 (Charach et al., 2011).

Prognosis and Outcomes

Alcohol use disorders are among the leading public health and safety concerns (World Health Organization [WHO], 2011). For example, youth who drink alcohol are more likely to experience low achievement, social issues, legal problems, and engage in risky sexual behavior (Meyers & Dick, 2010). In addition, because the adolescent brain is not fully developed, they are at an increased risk for changes in brain development that may have long-term effects (Meyers & Dick, 2010). Alcohol use over time is associated with a number of medical ailments including liver disease, cancer, and cardiovascular disease as well as the development of other mental health disorders such as antisocial personality disorder (Meyers & Dick, 2010). Though a significant health concern, the prognosis for individuals who seek treatment is quite promising (APA, 2013).

Smoking and Tobacco Use Disorders

Etiology

There is evidence for genetic and shared environmental influence on adolescent smoking (McGue, Elkins, & Iacono, 2000; Rende, Slomkowski, McCaffery, Lloyd-Richardson, & Niaura, 2005). Hopfer et al. (2003) found that the genetic contribution to tobacco use in adolescence ranges from modest to significant. Some studies report the heritability to be at 40% to 60% (McGue et al., 2000).

Estimates of shared environmental influences on high levels of smoking have been reported to range from 0.26 to 0.38 (McGue et al., 2000; Rende et al., 2005). However, research has showed that the environment has a stronger influence on tobacco in younger age groups (Hopfer et al., 2003). Household structure has been shown to affect adolescent smoking (Razaz-Rahmati, Nourian, Chizimuzo, & Okoli, 2012). For instance, the odds of an adolescent smoking in a single-parent household is 1.78 times greater than in a two-parent household. Further, exposure to secondhand smoke is associated with adolescent nicotine use (Razaz-Rahmati et al., 2012).

Associated Impairments and Comorbidities

There appears to be a relationship between cigarette use and childhood attention deficit hyperactivity disorder (ADHD). For example, individuals with ADHD have been found to be more likely to

take up cigarette smoking than individuals without ADHD (Looby, 2008; Wilens et al., 2011). Further, children with ADHD are upward of two times as likely to use nicotine than children without an ADHD diagnosis later during adolescence (Charach et al., 2011). Interestingly, the dimensions of ADHD appear to play a unique role in cigarette use. For instance, Molina and Pelham (2003) found that impulsivity/hyperactivity in childhood was related to an earlier onset for initial cigarette use, while childhood inattention was associated with heavier cigarette use.

Tobacco use is also associated with internalizing disorders (Cosci, Knuts, Abrams, Griez, & Schruers, 2010; Johnston et al., 2000; Sonntag, Wittchen, Hofler, Dessler, & Stein, 2000). For example, a longitudinal study of adolescents (ages 14–24) found that individuals with social phobia are more likely to develop dependence for nicotine later on (Sonntag et al., 2000). Further, Johnson et al. (2000) discovered that cigarette smoking might predict later anxiety disorders. Specifically, they found that heavy nicotine use was associated with anxiety disorders in emerging adulthood. In addition, adolescent smokers are at elevated risk of panic disorder and panic attacks. Adolescents who smoke 20 or more cigarettes per day are at elevated risk for panic disorder with agoraphobia during adolescence and early adulthood (Cosci et al., 2010).

Early nicotine use predicts later nicotine dependence as well as later abuse or dependence on alcohol and marijuana in young adulthood (Orlando, Tucker, Ellickson, & Klein, 2004; Palmer et al., 2009). Furthermore, nonsmokers are in better physical health than "triers" (smoke 1–2 cigarettes per year) of tobacco. Additionally, both of these groups report less limitation due to physical health than individuals who use tobacco more regularly. Nonsmokers and "triers" are more likely to graduate college than regular tobacco users (Orlando et al., 2004).

Course and Developmental Challenges

The majority of US adolescents experiment with tobacco use (APA, 2013). The first use of nicotine tends to occur at the age of 14 (Wittchen et al., 2007). By the age of 18, around 20% of individuals smoke at least monthly. Most of this, 20% become daily tobacco users (APA, 2013). Smoking in adolescence is related to alcohol use, peer influence, a history of depression, early sexual activity, and delinquency (APA, 2013; Lloyd-Richardson, Papandonatos, Kazura, Stanton, Niaura, 2002). It is common for adolescent smokers to also consume alcohol. The combination of early smoking and drinking predicts later substance abuse and dependence as well as further deviance (Orlando, Tucker, Ellickson, & Klein, 2005). Further, early nicotine use predicts later nicotine dependence as well as later abuse or dependence of alcohol and marijuana in young adulthood (Palmer et al., 2009). Symptoms of nicotine dependence occur in 20%–60% of adolescent smokers (Colby, Tiffany, Shiffman, & Niaura, 2000). Of every five adolescent users, between one and three are considered dependent on nicotine (APA, 2013).

Treatment

Adolescents who are impulsive find it especially difficult to quit smoking (Harris et al., 2014; Krishnan-Sarin et al., 2006). For example, one study found that better sustained attention (a measure of impulsivity) predicted later smoking cessation (Harris et al., 2014). Other research has focused on how to overcome factors that impede smoking cessation. For example, research has shown that contingency management (CM) (behavioral intervention that reinforces with immediate tangible rewards) is an effective strategy for helping these individuals quit smoking (Morean et al., 2015). Contingency management is far more effective with impulsive adolescents than cognitive behavioral therapy (CBT). For example, one study found that impulsive adolescents who received CM were 11.3 times more likely to be smoke-free at the end of treatment than individuals who were treated with CBT (Morean et al., 2015).

There is support for the use of school-based smoking cessation interventions (Pbert et al., 2011). For instance, one study examined the

effectiveness of a school nurse-delivered intervention. The intervention consisted of cognitive behavioral techniques and a counseling intervention. Results indicated that boys who received the treatment were nearly twice as likely to be abstinent after 3 months compared to the control group. However, this was not true at the 12-month mark. Therefore, this type of intervention is effective for short-term cessation in boys as well as short-term decline in amount and frequency in males and females (Pbert et al., 2011).

Some researchers believe that cessation treatments may need to include extended therapy and follow-up to be successful (McLellan, Lewis, O'Brian, & Kleber, 2000). Spurred by the poor smoking cessation rates of adolescents (Milton et al., 2004), Bailey et al. (2013) examined how effective extended treatment would be on adolescent smokers. Findings were positive for the success of extended treatments. The adolescents who were in the extended treatment group had a greater abstinence rate (21%) than the non-extended treatment (7%) group. Therefore, due to the higher abstinence rates for the extended treatment group, there is robust evidence for longer smoking cessation interventions with adolescents.

Prognosis and Outcomes

The leading cause of preventable mortality is nicotine dependence (WHO, 2008). While overall trends in cigarette smoking have declined since the 1980s, the number of adolescent smokers has not (US Department of Health and Human Services, 2012). Though adolescents will attempt to quit, their efforts may be in vain. Many will try to quit multiple times; however, cessation rates are considerably lower for adolescents compared to adults (Milton et al., 2004).

Outcomes for nicotine use consist of later dependence of other substances (Orlando et al., 2004; Palmer et al., 2009), poor physical health, a lack of educational attainment, and stealing (Orlando et al., 2004). Further, an important predictor of smoking cessation outcome is an adolescents' duration before having their first cigarette after waking. Research has shown that when adolescents have their first cigarette within 30 min of waking, they are less likely to have quit smoking at the end of a school-based quit program (Mercincavage, Branstetter, Muscat, & Horn, 2013).

Marijuana and Cannabis Use Disorders

Etiology

The heritability estimate for cannabis use is 45% for men and 39% for women (Verweij et al., 2010). Parental substance use disorder (SUD) status predicts adolescent boys frequency of cannabis use, the development of cannabis use disorder (CUD), and neurobehavior disinhibition (ND) (Feske et al., 2008). ND encompasses truncated executive functioning, little behavior control, and emotion dysregulation. Individuals with high ND become involved with peers who increase the likelihood of developing CUD. The interaction between ND and peer influences allows for the prediction of CUD outcomes with 84% accuracy. Further, boy's peer characteristics at age 16 predict CUD at age 19 and 22 (Feske et al., 2008).

Associated Impairments and Comorbidities

Marijuana use is associated with many negative outcomes. For example, marijuana users show an increase in sexual activity as well as risky sexual behaviors (Anderson & Stein, 2011). Further, early onset of cannabis use is associated with poor performance on sustained attention, impulse control, and executive functioning (Fontes et al., 2011).

Unfortunately, there are common comorbid externalizing disorders with marijuana use. For instance, there is an association between mari-

juana use and conduct disorder symptoms (Wymbs et al., 2014). In addition, childhood ADHD is a significant predictor of the later development of cannabis use disorder (Charach et al., 2011). However, some studies have shown that the increased likelihood for substance use disorders (SUDs) and ADHD with marijuana dependence only holds true for females (Roberts et al., 2007).

In terms of internalizing disorders, there is also comorbidity. There is an association between cannabis use (CU) as well as cannabis use disorder (CUD) and depression symptoms (Brook, Brook, Zhang, Cohen, & Whiteman, 2002; Wittchen et al., 2007). Researchers have found an association between CUDs, mood disorders (including bipolar disorders), generalized anxiety disorder (GAD), and separation anxiety. Panic anxiety and separation anxiety are associated with CU as well as CUD (Wittchen et al., 2007). It has been found that 22% of individuals with CUD have previously diagnosed anxiety disorders and 37% have formerly diagnosed affective disorders (Hofler et al., 1999). Others have found that CU puts individuals at an increased risk of psychotic symptoms (Henquet et al., 2004).

Having a mental illness can also predict CU and CUDs. For example, dysthymia, panic anxiety, conduct problems, and ADHD predict later CU. Further, bipolar disorders and depressive disorders predicted CU and CUD. Taken together, mood disorders are a significant predictor for CU and CUD. However, the influence of anxiety disorders is inconstant. Finally, before individuals had a CUD, 53.7% of people suffered previously diagnosed mental or substance use disorder. Of those that had a prior disorder, 68.8% had an alcohol use disorder, while 63.4% had a nicotine use disorder (Wittchen et al., 2007).

Course and Developmental Challenges

In the United States, cannabis is the most frequently used illicit drug by adolescents and emerging adults. The frequency of CUD is almost equal that of the combined incidence of other illicit substance use disorders (SAMHSA, 2009). Further, cannabis is typically the first substance adolescents experiment with (Parritz & Troy, 2014). Initial use of cannabis occurs around the age of 15 (Wittchen et al., 2007). New marijuana users tend to be between the ages of 12 and 17 (SAMHSA, 2004). Around 14% of adolescent-onset marijuana use will result in cannabis dependence, whereas adult-onset users develop cannabis dependence at half that rate (Chen, Kandel, & Davies, 1997). In addition, early onset of cannabis use is also associated with the development of other substance use disorders, mental disorders, and the failure to meet education milestones such a completion of high school, enrolment in a university, and obtaining a degree (APA, 2013; Horwood et al., 2010; McCaffrey, Pacula, Han, & Ellickson, 2010; Parritz & Troy, 2014).

Furthermore, adolescents with this disorder also typically experience school-related problems consisting of a drop in grades, truancy, and decreased interest in school activities and outcomes. Some studies suggest that marijuana use is associated with reductions in executive functioning, related to attention, memory, and learning. Other studies suggest that marijuana is associated with lower rates of school achievement and graduating from high school (APA, 2013; Parritz & Troy, 2014). Adolescent users are apt to show deviations from mood stability, energy levels, and eating patterns. Cannabis use disorder tends to develop over a long period of time. However, the progression appears to be faster in adolescents, especially for those with conduct problems (APA 2013; Parritz & Troy, 2014). Fortunately, among adolescents, marijuana use has decreased from 27% in 1999 to 20% in 2007 (Johnston, O'Malley, Bachman, & Schulenberg, 2007).

Treatment

Treatment of cannabis use has been shown to be effective with cognitive behavioral therapy

(CBT) interventions (Dennis et al., 2000; Waldron et al., 2001). Dennis et al. (2000) found that there was a significant decrease in the percentage of substance use from pretreatment to a 3-month follow-up with CBT treatment. Further, Waldron et al. (2001) tested the effectiveness of CBT, family therapy, combined individual and family therapy, and group intervention on adolescents. Results indicated that while all of the interventions showed a degree of efficacy, there was a significant reduction in use from pretreatment to 4 months with family therapy alone and combined individual and family therapy. In addition, a significantly greater amount of adolescents were at the minimal use level after family, combined, and CBT. Further, from pretreatment to 7 months, there was a significant drop in use for combined and group intervention. Finally, family, combined, and group therapy significantly impacted the number of adolescents at the minimal use level after 7 months.

Another effective treatment consists of Adolescent Community Reinforcement Approach paired with Assertive Continuing Care (A-CRA/ACC). A-CRA is a 3-month individualized behavioral treatment plan. This treatment starts with an evaluation of the disorder and triggers. ACC is a support for adolescents consisting of skills training. It also aids adolescents in keeping social environments that endorse non-substance use (McGarvey et al., 2014). McGarvey and colleagues (2014) found that the A-CRA/ACC treatment was effective with their adolescent sample. At the end of 12 months, greater than two-thirds (68.7%) of the adolescence had quit. However, for those that were unable to quit, they were the heaviest users. This suggests that this treatment may be most effective with moderate cannabis users (McGarvey et al., 2014).

Research has shown that adolescents tend to have similar substance use problems as their peers (Becker & Curry, 2014). Becker and Curry (2014) tested whether this was due to peer socialization (adolescents adopt the beliefs and attitudes of their peers) or peer selection (adolescents choose peers with comparable beliefs and attitudes). They found that adolescent marijuana use was influenced more by peer selection. Therefore, when treating cannabis use disorder, it is relevant to help adolescents choose prosocial peers (Becker & Curry, 2014).

Prognosis and Outcomes

The purchasing behavior of adolescents has been shown to correlate with their prognosis. Osilla et al. (2014) found that individuals who purchase marijuana tend to use marijuana more frequently than users who do not purchase. Purchasers are also associated with spending time with peers who use marijuana and are more likely to have a substance use disorder than non-purchasers of marijuana.

Marijuana use trajectories have been found to be different for different genders when studying African American adolescents. Juon, Fothergill, Green, Doherty, and Ensminger (2011) studied a cohort of African American youth through the age of 32. They found that persistent male users were at the highest risk of having a substance use disorder, being incarcerated and being unmarried, and have a high incidence of major depressive disorder (MDD). Persistent female users were at an elevated risk of having a substance use disorder, being incarcerated, and having MDD. Further, abstainers and desistance (used marijuana but desist by adolescence or adulthood) individuals were more likely to be married than the heavier, later users.

Other Substance Use Disorders: Phencyclidine, Opioid, Sedative, Hypnotic, Anxiolytic, Stimulant

Etiology

Relatives of individuals with one or more SUDs (consisting of opioids, cocaine, cannabis, and alcohol) have been found to be around eight times more likely to suffer from any kind of SUD. It has been stated that a family history of SUDs is one of the biggest risk factors for developing a drug disorder (Merikangas et al., 1998). Heritability estimates for hallucinogens have been reported at 49%, 52% for opioids, 60% for sedatives, and 21% for stimulants (Kendler,

Kirkowski, & Prescott, 1999). Recently, researchers have found that the genetic risk for cocaine and stimulant SUD is similar across substances. Therefore, an individual who has high genetic risk for dependence of stimulants would likely have the same risk for becoming dependent on cocaine (Kendler, Jocobson, Prescott, & Neale, 2003; Ystrom, Reichborn-Kjennerud, Neale, & Kendler, 2014).

Environmental influences have also been implicated in SUDs. Shared experiences can increase the likelihood of illicit substance use in twins. These experiences also relate to general risk rather than risk for a specific substance. Therefore, it is unlikely that shared experiences form substance-specific risk (Kendler et al., 2003; Ystrom et al., 2014). Kendler and colleagues (2003) found that environmental experiences unique to an individual principally determine whether predisposed individuals will use or misuse one class of psychoactive substance rather than another. However, while there are environmental risk factors that are unique to individuals, these risk factors do not uniquely determine abuse or dependence of specific substances (Ystrom et al., 2014).

Associated Impairments and Comorbidities

Charach et al. (2011) found that an ADHD diagnosis in childhood is predictive of psychoactive SUD during emerging adulthood. Inhalant abuse is associated with a permanent decline in cognitive performance and heart arrhythmias. Long-term inhalant users are at risk for depression, anxiety, bronchitis, asthma, and sinusitis (APA, 2013). MDMA (ecstasy) is associated with damage to serotonin-containing neurons, impairment in memory, sleep disturbance, white matter reduction, and damage to axons (APA, 2013). There is also an association between ecstasy use and mental disorders. Frequently, first use of ecstasy occurs after the onset of mental disorders (Lieb, Schuetz, Pfister, von Sydow, & Wittchen, 2002).

Opioid use is associated with dry mouth and nose and slowing of gastrointestinal activity, and for those who inject opioids, veins can become sclerosed, and edema may develop (APA, 2013). There is a significantly lower incidence of ADHD in the populations that use cocaine as their drug of choice (van Emmerik-van Ootmerssen et al., 2012). Sedative, hypnotic, or anxiolytic use disorder is associated with accidents, interpersonal problems, and hindrance of school performance, and high doses can be lethal (APA, 2013).

Course and Developmental Challenges

Overall, illicit drug use is not common among adolescents. The prevalence of illicit drug dependence is around 3.3% (Weinberg et al., 1998). Inhalant use reaches its height between seventh and ninth grade. Use is most serious for adolescents who have restricted access to alcohol or other drugs. About 0.4% of adolescents (ages 12–17) meet the criteria for inhalant use disorder. The peak of inhalant use is between seventh and ninth grade, with continued use more frequent in boys (APA, 2013; Parritz & Troy, 2014).

The use of cocaine among twelfth graders is 0.2% (Johnston et al., 2001). Rates are highest among 18–29-year-olds (0.6%). Frequency of use for males (0.4%) is higher than among females (0.1%). Unfortunately, cocaine has the highest probability for the transition from regular use to abuse/dependence. Long-term crack cocaine use is associated with PTSD, antisocial personality disorder, and suicide risk. Tobacco, alcohol, cannabis, amphetamine, and cocaine dependence are also associated with lifetime use of crack cocaine (Narvaez et al., 2014). Cocaine use can cause cardiovascular, respiratory, and neurological impairments. Amphetamines have similar usage rates among 12–17-year-olds (0.2%). The rate for females (0.3%) is greater than that of males (0.1%). Intravenous stimulant use occurs more frequently in males; the ratio of male to female is 3:1 or 4:1. However, rates are

fairly equal among non-injection users (APA, 2013; Parritz & Troy, 2014).

The use of the hallucinogen among high school students has dropped from 13% in 2001 to 8% in 2007 (Johnston, O'Malley, Bachman, & Schulenger, 2009). Furthermore, among twelfth graders the prevalence of use is around 3.5% (Johnston et al., 2001). Phencyclidine (PCP) use increases with age. For 12–17-year-olds the prevalence is 0.3% of the population. Use of ecstasy among high school students has dropped nearly in half from 2003 (11%) to 2007 (6%) (Johnston et al., 2009). This use disorder is one of the least common diagnoses of all the substance use disorders. The prevalence is around 0.5% among 12–17-year-olds. LSD can cause long-term complications, such as flashbacks (APA, 2013; Parritz & Troy, 2014).

Adolescents (ages 12–17) who meet the criteria for opioid use disorder is approximately 1.0% in the United States. Rates are higher for males than females about 1.5:1 for opioids other than heroin and 3:1 for heroin. Problems with opioid use tend to occur in the late teens to early 20s. Heroin is correlated with the highest probability for the transition between repeated use and regular use (APA, 2013; Parritz & Troy, 2014).

Sedative, hypnotic, or anxiolytic use disorder is typically diagnosed in individuals who begin use in teens or 20s and whose use escalates. While illicit drug use among adolescents has dropped, the high use of nonmedical prescription and over-the-counter (OTC) medication (including pain relievers, tranquilizers, stimulants, and depressants) has not wavered (Meyers & Dick, 2010). The recent increase in the use of prescription drugs is second only to the initiation rates for cannabis (SAMHSA, 2011). Pain medications, depressants, and stimulants are the most commonly abused prescription drugs (National Institute on Drug Abuse [NIDA], 2012). In 2011, 10% to 15% of high school seniors used prescription drugs for nonmedical purposes (NIDA, 2012; SAMHSA, 2011). The prevalence of this use disorder for 12–17-year-olds is around 0.3%. The rate for females (0.4%) is greater than for males (0.2%) (APA, 2013).

Treatment

Grella et al. (2001) found that adolescent users of cocaine, hallucinogens, and other stimulants did not respond well to short-term inpatient (STI) or outpatient drug-free (ODF) programs. While the majority of adolescents who seek treatment use alcohol or marijuana, there needs to be more focus on illicit drugs. Therefore, treatment programs need to better address the needs of those who use illicit drugs.

Treatment effectiveness of methadone-maintained individuals has been examined in high-baseline opiate and cocaine users (Chutuape, Silverman, & Stitzer, 1999). Take-home incentives can help reduce drug use. The conditions that promote success are high doses of methadone, immediate dispersion of take-home rewards following abstinence, and a focus on only a few illicit drugs (Chutuape et al., 1999).

Prognosis and Outcome

There are conditions under which treatment for opioid, cocaine, and hallucinogen use have been found to be effective (Chung & Martin, 2005; Weiss et al., 2014). For in- and outpatient programs, teens were more likely to transition to a less severe class of usage than to move to a more severe class over the period of a year. Also, those in the mild and severe class had a high probability of moving to the lowest severity class after 1 year (Chung & Martin, 2005). Heroin users who have received drug counseling are more likely to become abstinent from opioids than users who have received only medical management. This only holds true if users adhere to treatment and receive adequate counseling. This emphasizes the importance of treatment adherence (Chung & Martin, 2005).

There is a link between substance use and suicide (Wong, Zhou, Goebert, & Hishinuma, 2013). Heroin use in particular has been shown to be associated with suicide and suicide ideation. Researchers have found that adolescents reporting heroin use also report the highest odds of sui-

cide ideation, suicide planning, suicide attempts, and severe suicide attempts over the past year. Methamphetamine use is also highly linked with reporting suicide ideation, plans, attempts and severe attempts compared to nondrug users. These odds are followed by the use of steroids, cocaine, inhalants, hallucinogens, and ecstasy (Wong et al., 2013).

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Part XII

Externalizing Disorders: Relationship Problems

Parent-Child and Sibling Relational Problems

29

Jessica G. Linscott and Leslie D. Leve

In the *Diagnostic and Statistical Manual of Mental Disorders* (5th ed., *DSM-5*; American Psychiatric Association [APA], 2013), parent-child and sibling relational problems are considered within a category of other conditions that may be a focus of clinical attention, rather than comprising a specific diagnosis or mental disorder. Thus, this chapter differs somewhat from the preceding chapters in this volume due to the lack of a specific diagnosis for parent-child and sibling relational problems. Nonetheless, relational problems are clinically significant issues that may impact other diagnoses and treatment plans. In this chapter, we present a description of parent-child and sibling relational problems, review their history in the *DSM*, and summarize the research about them with a focus on etiology, symptom presentation, associated impairments and comorbidities, course and developmental challenges, and treatment and outcomes. We incorporate a discussion of the role of family dysfunction as a broad risk factor in the development of parent-child and sibling relational problems, although abuse and neglect are not a primary focus of this chapter and have their own V-codes in the *DSM-5* (APA, 2013). We begin by presenting a description of parent-child relational problems and sibling relational problems and subsequently review the research in the areas noted above. Because the associated etiologies, impairments, treatments, and outcomes are interrelated, our research review for the two types of relational problems is integrated.

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Problems Related to Family Upbringing: Definitional Issues

In the *DSM-5*, "parent-child relational problem" and "sibling relational problem" are found within the category of "Problems Related to Family Upbringing." Both are a specific type of "relational problem" specified in the *DSM-5*, categorized within "Other Conditions That May Be a Focus of Clinical Attention." These conditions are not considered mental disorders or diagnoses; rather, they are considered to be problems that may be relevant for clinical treatment or may affect the diagnosis and course of an individual's mental disorder. According to the *DSM-5*, mental disorders are limited to clinically significant behavioral or psychological syndromes or patterns residing within an individual, whereas relational problems occur between or among individuals (APA, 2013).

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Although not a mental disorder, the inclusion of a relational problem as identified with a V-code within a *DSM-5* diagnosis provides further contextual information regarding pertinent relational issues and allows the clinician to adequately document the full range of a patient's issues relevant to assessment, diagnosis, and treatment. Documentation of a relational problem may or may not reflect the primary reason for a patient's visit, but may be relevant for assessment or treatment. While the *DSM-5* does not provide specific diagnostic criteria for assessment of the appropriate use of these V-codes, it does provide brief narrative descriptions, which are summarized in this chapter.

According to the *DSM-5*, relational problems reflect significant clinical distress or disturbance of key relationships such as the parent-child or intimate adult partner relationship. Relational problems have "a significant impact on the health of the individuals in these relationships. These relationships can be a health promoting and protective, neutral, or detrimental to health outcomes" (APA, 2013, p. 715). Parent-child relational problems, while potentially impairing functioning in the individual or family, represent non-abusive relationship problems when considered in the context of a relational continuum. As such, the *DSM-5* does not include defined criteria or symptom presentation for relational problems.

Child maltreatment, another condition that may be a focus of clinical attention, is given its own category in the *DSM-5* and represents the more extreme, abusive end of disordered parenting and parent-child relational problems (Heyman & Slep, 2006). Indeed, "abuse and neglect" is given a section within "Other Conditions That May Be a Focus of Clinical Attention" in the *DSM-5*, with subcategories for "child maltreatment and neglect" that include child physical abuse, child sexual abuse, child neglect, and child psychological abuse, whether confirmed or suspected (ICD-10-CM codes 995.54, 995.53, 995.52, and 995.51, respectively). While a complete description of child maltreatment and neglect is beyond the scope of this chapter, there is a wide body of literature documenting the detrimental developmental effects of child maltreatment for the affected child and associated negative health outcomes (for reviews, see Brown, Fang, & Florence, 2011; Twardosz & Lutzker, 2010).

Parent-Child Relational Problem (V61.20)

According to the *DSM-5*, the category of "Parent-Child Relational Problem" should be used when the focus of clinical attention "is to address the quality of the parent-child relationship or when the quality of the parent-child relationship is affecting the course, prognosis, or treatment of a mental or other medical disorder" (APA, 2013, p. 715). The term *parent* is defined as a primary caregiver to the child; this caregiver may be a biological, adoptive, or foster parent or other relatives who fulfill a parenting role. Associated impairment in cognitive, affective, or behavioral functioning is typically associated with a parent-child relational problem (APA, 2013). Examples in the cognitive realm include negative attributions of the others' intentions, hostility or scapegoating, and unwarranted feelings of estrangement. Examples of impairment to affective functioning include feelings of sadness, apathy, or anger regarding the other individual. Examples of behavioral problems include: parental overprotection; inadequate parental control, supervision, and involvement with the child; excessive parental pressure; avoidance without the resolution of problems; and the escalation of arguments to threats of physical violence (APA, 2013). Further, the description of the problem calls for the clinician's attention to developmental and cultural factors of the child and family context that may impact the inclusion of the problem within a *DSM-5* diagnosis.

Sibling Relational Problem (V61.8)

There are many parallels between the definition and course of the sibling relational problem and that of the parent-child relational problem. A *DSM-5* category appropriate for use with chil-

dren or adults, "Sibling Relational Problem," is used when the focus of clinical attention or treatment is "a pattern of interaction among siblings that is associated with significant impairment in individual or family functioning" (APA, 2013, p. 716). It may also be used if the sibling problem is associated with the development of clinical symptoms in one or more siblings, if a sibling relational problem is affecting the course, prognosis, or treatment of a sibling's mental or medical disorder. The *DSM-5* specifies that a sibling can include full, half-, step-, foster, and adopted siblings. Specific examples of sibling relational problems (e.g., sibling rivalry or aggression) are not specified. As with other listed relational problems related to family upbringing (e.g., "parent-child relational problem," "upbringing away from parents," and "child affected by parental relationship distress"), sibling relational problems can be significant because sibling relationships can serve as key developmental and adult relationships, impacting the health and well-being of affected individuals. The *DSM-5* specifies that a sibling relational problem may be the primary reason that an individual seeks care or may be a contributing factor that affects an individual's other mental or medical disorder.

History of Parent-Child and Sibling Relational Problems in the *DSM*

The earliest mention of the "Parent-Child Relational Problem" and "Sibling Relational Problem" categories appeared in the third edition of the *Diagnostic and Statistical Manual of Mental Disorders* (3rd ed., *DSM-III*; American Psychiatric Association [APA], 1980). A V-code category labeled "Parent-Child Problem" was described briefly as of use when a focus of treatment involves a parent-child problem that is not due to a mental disorder of the parent or child, with child abuse serving as the sole example. Thus, in the *DSM-III*, parent-child relational problems and child maltreatment were not yet assigned separate categories. Elsewhere in the *DSM-III*, it was noted that childhood conditions that did not meet criteria for a diagnosis of a mental disorder might be captured with a V-code. The text specified that child abuse did not warrant a diagnosis as a mental disorder of the parents as it was not necessarily associated with any mental disorder and/or could be symptomatic of a variety of disorders (APA, 1980). Child abuse, according to the *DSM-III*, could also adequately serve as an example of "Other Specified Family Circumstances."

Also in the *DSM-III*, a category labeled "Other Specified Family Circumstances" was briefly described therein as of use when a focus of treatment or clinical attention was a family circumstance not due to a mental disorder and not better described as a parent-child or marital problem (APA, 1980). Sibling rivalry served as one of two examples of such a circumstance, along with interpersonal difficulties with an aged in-law. This was the only inclusion of concerns that would subsequently be categorized as sibling relational problems.

A revised version of the third edition of the *DSM* (3rd ed., rev., *DSM-III-R*; American Psychiatric Association [APA], 1987) provided a nearly identical definition of parent-child problem, with the exception of the examples provided. In this case, an example provided is a "conflict between a mentally healthy adolescent and her parents about her choice of friends" (APA, 1987). No mention of child maltreatment is found within the *DSM-III-R*; the term *child abuse* in the glossary refers the reader to *parent-child problem*. No changes were made to the "Other Specified Family Circumstances" category in the *DSM-III-R*, and as such sibling relational problems were still subsumed under this broad category.

With the publication of the fourth edition of the *DSM* (4th ed., *DSM-IV*; American Psychiatric Association [APA], 1994) and its subsequent revision (4th ed., rev., *DSM-IV-R*; American Psychiatric Association [APA], 2000), parent-child relational problem was given as a specific V-code of the subcategory of relational problems within "Other Conditions That May Be a Focus of Clinical Attention." This was the first time that specific relational problems were named and grouped together. Relational problems were noted to be included within Axis I of a diagnosis

if the relational problem was the primary clinical focus or to be listed on Axis IV if a relational problem was present but was not the central focus of clinical attention (APA, 1994). With the *DSM-IV* came a more detailed description of the parent-child relational problem, which specified the use of the category when the focus of clinical attention was a "pattern of interaction between parent and child… that is associated with clinically significant impairment in individual or family functioning or the development of clinically significant symptoms in parent or child" (APA, 1994, p. 681). Examples included impaired communication, overprotection, and inadequate discipline. The *DSM-IV-TR* (APA, 2000) introduced a separate relational category section, "Problems Related to Abuse or Neglect," specifying separate categories for physical abuse, sexual abuse, and neglect of children, thus giving child maltreatment separate and distinct categories from other relational problems for the first time.

A separate category for sibling problems first appeared in the *DSM-IV* and again within the *DSM-IV-TR* as "Sibling Relational Problem," also a V-code. It is described as a category in which "the focus of clinical attention is a pattern of interaction among siblings that is associated with clinically significant impairment in individual or family functioning or the development of symptoms in one or more of the siblings" (APA, 1994, p. 681).

The *DSM-5* marked the first time that a definition of the term *parent* was included in the category description. In addition, the description of the problem in the *DSM-5* also marked the first inclusion of the importance of the role of developmental needs and culture of the child for clinical consideration. The *DSM-5* more explicitly describes domains of impaired functioning as a result of parent-child relational problems, including potential behavioral, cognitive, and affective impacts. Both "Parent-Child Relational Problem" and "Sibling Relational Problem" are included as specific categories of relational problems related to family upbringing in the *DSM-5*. Other related categories of problems related to family upbringing include two new categories that appear within the *DSM-5*, "Upbringing Away from Parents" and "Child Affected by Parental Relationship Distress." "Upbringing Away from Parents" is a V-code that captures situations in which the main focus of clinical attention is related to issues of child rearing away from the parents (e.g., state custody kin care or foster care, living in an orphanage or group home, or non-court-mandated living with nonparental relatives or friends). "Child affected by parental relationship distress," while a condition that may be associated with parent-child or sibling relational problems, is specified when the focus of clinical attention is the detrimental effects of parental relationship distress or discord on a child. Parental alienation, a process by which a child is manipulated by a parent into preferring one parent to another (Bernet, 2008), while not included specifically as a "Relational Problem" category in the *DSM-5* or as an example of parent-child relational problems, could serve as an example of the "Child Affected by Parental Relationship Distress" category or as a type of parent-child relational problem associated with parental relationship distress.

"Other Problems Related to Primary Support Group" represents another category of relational problems in the *DSM-5*, within "Other Conditions That May be A Focus of Clinical Attention." The specific subcategories contained therein include "Relationship Distress with Spouse or Intimate Partner," "Disruption of Family by Separation or Divorce," "High Expressed Emotion Level Within Family," and "Uncomplicated Bereavement." While the presence of one relational problem in a *DSM-5* diagnosis does not preclude another (i.e., there can be present both parent-child relational disorder and high expressed emotion level within a family), the clinician is encouraged to highlight the problem most associated with presenting clinical distress and impairment in the primary V-code diagnosis, with other relational problems identified as supplemental information or as documentation aid (APA, 2013).

Overall, a comparison of the inclusion of parent-child and sibling relational problems across versions of the *DSM* reflects increasing attention to the issues, more thorough description of the categories, greater use of examples,

and the provision of additional guidance for clinicians regarding the appropriate use of the categories. Still, many researchers have urged even greater attention to the role of relational processes and problems for individual and family systemic functioning in the diagnostic texts of the APA and have argued for the inclusion of specific criteria for the assessment of relational disorders (e.g., Beach & Kaslow, 2006; Beach, Wamboldt, Kaslow, Heyman, & Reiss, 2006a; Wamboldt & Reiss, 2006). Identifying the need for operationalization of constructs and specific assessment instruments, Heyman et al. (2009) proposed specific diagnostic criteria for the assessment of relational problems, including partner relational problems, parenting problems, and child physical abuse, and Wamboldt and Cardaro (2012) reviewed proposed diagnostic criteria for caregiver-child relational problems. Although not incorporated into the *DSM-5* as a relational disorder, the clear trend for the increased focus on relational processes over time across versions of the *DSM* may be a sign that specific relational problems could rise to the level of an independent disorder in future *DSM* volumes.

Etiology of Parent-Child and Sibling Relational Problems

Although a substantial body of literature documents the etiology and effects of child maltreatment, there is more limited research on less extreme family-based "relational problems" as specified in the *DSM-5*. However, research in the area of relational problems has begun to burgeon over the past decade (Beach, Wamboldt, Kaslow, Heyman, & Reiss, 2006b). Current challenges for research regarding relational problems include the lack of defined criteria and symptom presentation provided by the *DSM*, making operationalization of variables and research regarding the valid and reliable assessment and treatment of the *DSM*-defined relational problems difficult (Slep & Tamminen, 2012; Wamboldt & Cardaro, 2012).

Nonetheless, while not specific to the "Parent-Child Relational Problem" or "Sibling Relational Problem" codes in the *DSM-5*, an extensive body of literature exists reflecting the crucial role of family processes, parenting styles, and parenting practices as they impact the parent-child relationship, sibling relationships, and developmental outcomes for the child, including impairment in cognitive, affective, and/or behavioral functioning. Children at the highest risk for negative family dynamics often show comorbid internalizing and/or externalizing disorders (Granic & Lamey, 2002). This process can be reciprocal, as a child's internalizing and/or externalizing symptoms may negatively affect the quality of the parent-child relationship dynamic; however, a negative parent-child dynamic may also contribute to the development of internalizing or externalizing symptoms in the child (Bell & Chapman, 1986; Harold et al., 2013; Lansford et al., 2011).

The coercion process, a family dynamic associated with externalizing and antisocial behaviors in children (Snyder, Reid, & Patterson, 2003), is highly relevant to the etiology of parent-child and sibling relational problems. Within the coercive cycle, a child or adolescent learns that by engaging in aversive behaviors, a parent's demands can be avoided or manipulated such that the child does not perform the requested behavior (Dishion & Stormshak, 2007; Patterson, 1982). For example, a child may learn that by escalating negative behavior from whining to a full tantrum, a parent may give in and provide the child what they desire or dismiss a previous request made of the child. In this way, the child learns that increasing negative behavior can lead to achievement of his/her own goals, which run contrary to parental demands. The parent, worn down by such behaviors, may withdraw or instead escalate emotionally, leading to further parenting problems and parent-child relational discord. This coercive cycle is associated with oppositional and antisocial behavior in childhood (Shaw et al., 1998; Patterson, Reid, & Dishion, 1992), as well as with antisocial behavior in adolescence (Dishion & Andrews, 1995).

Similarly, coercive dynamics can occur within the sibling relationship. A negative mother-child relationship is predictive of sibling conflict, which then predicts the child's engagement in antisocial behavior (Criss & Shaw, 2005).

In addition, ineffective parenting can facilitate a younger siblings' exposure to an older siblings' deviant peers and activities, providing opportunities for older peers or siblings to train younger siblings in antisocial activities (Snyder, Bank, & Burraston, 2005). Further, siblings can reinforce each other's antisocial tendencies and expose each other to risk-taking behaviors such as substance use, delinquency, and other delinquent peers (Bricker et al., 2006; Snyder et al., 2005). The family can thus serve as a training ground for the development of clinical problems, with coercive cycles that begin in the family (parent-child and sibling interactions) teaching youth to develop a coercive interactional style, which then translates to the peer and school realm (Patterson, 1982; Patterson, Capaldi, & Bank, 1991). While identification of the coercive cycle may pose challenges for the clinician, effective assessment of such processes may aid them in determining appropriate treatment for parent-child or sibling relational problems.

Symptom Presentation

The presentation of symptoms of parent-child or sibling relational problems may not be readily identifiable to the clinician, as the relational problem exists within the relationship of two or more individuals, rather than as symptoms of a disorder within either party. However, parent-child and sibling relational problems may also be the product of associated distress or environmental factors due to another mental or medical disorder found within a member of the family system. The clinician must assess for additional disorders present among individuals and within the family context in order to create a full, contextual diagnostic picture.

No specific symptoms are listed in the *DSM-5* for either parent-child or sibling relational problems. However, examples of problems that may be indicative of parent-child or sibling relational problems are presented in the *DSM-5* and are described in the existing research literature on the subject (e.g., Beach et al., 2006b; Slep & Tamminen, 2012; Wamboldt & Cardaro, 2012). According to the authors, child behaviors that may be indicative of relational problems include:

- High levels of negativism and/or disapproval
- Noncompliance and/or engaging in aversive behaviors to avoid parental requests

Parental behaviors that may be indicative of relational problems include:

- Overinvolvement or underinvolvement
- Overprotection or underprotection
- Harsh and critical parenting
- Overreactive anger
- Inadequate, harsh, and/or inconsistent discipline
- Rigidity
- Hostility toward child
- Rejection of child
- Exertion of high levels of control
- Extremely high or low levels of expressed emotion in the affective style of the parent
- Developmentally unrealistic expectations of child

Conversely, functional parenting is more likely marked by:

- Appropriate discipline
- Parental warmth
- Appropriate autonomy granting of the child
- Adequate parental monitoring and supervision

The factors presented above are best considered along a continuum of functional to dysfunctional parenting, rather than as dichotomous characteristics of parenting. As such, higher levels of the child or parental behaviors described above may serve to alert the clinician that an assessment of the family system dynamics may be useful in indexing risk for relational problems and their comorbid impairments. No specific examples are provided for sibling relational problems, but due to the family systems nature of the etiology of sibling relational problems, the same parenting descriptors listed above are relevant for sibling relational problems. Differential treatment of siblings and children's perceptions of fairness can also have specific effects on sibling adjustment (Brody, Stoneman, & McCoy, 1992; Kowal, Krull, & Kramer, 2004; McHale, Updegraff, Jackson-Newsom, Tucker, & Crouter, 2000; Tamrouti-Makkink, Dubas, Gerris, & van Aken, 2004).

Associated Impairments and Comorbidities

A range of other diagnoses may be comorbid with parent-child or sibling relational problems. In fact, the cornerstone of the definition of parent-child relational problems is that the quality of the parent-child relationship is affecting the course, prognosis, or treatment of a mental or other medical disorder; similarly, the definition of sibling relational problems emphasizes significant impairments in individual or family functioning. Thus, it is likely that parent-child and sibling relational problems occur in tandem with individual disorders and may come to the attention of the clinician in the course of treating one of more primary disorders. The most prevalent comorbid diagnoses and impairments are described below.

Child Oppositional Defiant Disorder, Conduct Disorder, and Attention Deficit Hyperactivity Disorder There is significant research evidence that oppositional defiant disorder (Chap. 31), conduct disorder (Chap. 32), and attention deficit hyperactivity disorder (ADHD; Chap. 26) are often comorbid with parent-child and sibling relational problems. Relational processes between parent and child are considered fundamental to the development and maintenance of conduct disorder and early-onset conduct problems (Beach et al., 2006b; Patterson, 1982), which may be symptomatic of parents' lack of reinforcement of appropriate behavior. Coercive processes between parents and children and other parent characteristics may serve to maintain inappropriate behaviors in children (Kazdin, 1995; Patterson, 1982; Patterson, DeGarmo, & Knutson, 2000). Correlates of negative parenting practices, including inadequate supervision, inconsistent and coercive discipline, a hostile and punitive style, as well as child maltreatment, are associated with conduct problems in children (Lahey, Miller, Gordon, & Riley, 1999; Patterson & Forgatch, 1995; Patterson & Stouthamer-Loeber, 1984). Further, a child diagnosed with oppositional defiant disorder, conduct disorder, or ADHD may present great challenges to effective parenting and lead to unusual strain on the parent-child relationship. Children with ADHD pose more challenges to parents and other adult figures than children without this disorder; problematic parent-child relations have also been found to predict symptoms of ADHD (Johnston & Mash, 2001; Lifford, Harold, & Thapar, 2008). Parents of children with ADHD are also more likely to report more negative-reactive and fewer positive parenting strategies on a daily basis than are parents of control (non-ADHD) children (Johnson, 1996). Further, maternal psychosocial functioning tends to be lower among mothers of children with ADHD and among fathers with children diagnosed with ADHD and oppositional defiant behavior (Johnson, 1996). Overall, it is clear that oppositional defiant disorder and conduct disorder can co-occur with parent-child and sibling relational problems and that children with either diagnosis or with a diagnosis of ADHD can be taxing to the family system, leading to relational disruptions and problems.

Childhood Anxiety Disorders Childhood anxiety disorders such as separation anxiety disorder (Chap. 9), generalized anxiety disorder (Chap. 11), or panic disorder (Chap. 9) may also be comorbid with parent-child or sibling relational problems, as parenting and parent-child interactions play an important role for the development childhood anxiety (Bögels & Brechman-Toussaint, 2006; Rapee, Schniering, & Hudson, 2009; Wood, McLeod, Sigman, Hwang, & Chu, 2003). Overprotective or over-controlling and harsh, critical, or negative styles (e.g., lack of warmth and acceptance) of parenting are associated with the development of childhood anxiety disorders; this association is largest and most consistently found for overprotective parenting (Rapee, 1997; Wood et al., 2003). However, more experimental research is needed to determine causal and directional effects of the associations between parenting and childhood anxiety, as parent and child behavior is often reciprocal and mutually influenced (Bögels & Brechman-Toussaint, 2006; Hudson & Rapee, 2004). For example, symptoms of anxiety in the child may lead to overprotective parenting behaviors, and overprotective parenting behav-

iors may further elicit anxious behaviors in the child. Parents of children with separation anxiety disorder may unintentionally promote childhood anxiety through overprotection, excessive reassurance, and aversive parent-child interactions (Eisen, Engler, & Geyer, 1998). Additionally, gene-by-environment interaction effects for the development of childhood anxiety is an area of needed future research, as factors such as child temperament and genetic predisposition may interact with the parenting environment to shape the development of childhood anxiety (Natsuaki et al., 2013; Rapee et al., 2009).

Other Childhood Disorders Other childhood disorders that are likely to be comorbid with parent-child and/or sibling relational disorders include mood disorders (Chaps. 7 and 8) and substance use disorders (Chap. 36). Problematic parent-child relations have been found to predict risk for depression (Blatt & Homann, 1992), with childhood depression associated with negative and critical parenting (Rapee, 1997). Additionally, substance use disorders in children have been found to be associated with parental psychopathology (Buu et al., 2009) and may be comorbid with parent-child relational problems related to parenting styles (Cohen & Rice, 1997). Among trauma- and stressor-related disorders, reactive attachment disorder (Chap. 16) in children may also be comorbid with severe parent-child relational problems and child abuse or neglect (Zeanah et al., 2004), as it is associated with a history of insecure attachment to caregivers and unstable or problematic caregiver relationships.

Parental Diagnoses When considering potential comorbidities, the clinician ought also to consider parental psychopathology that may be present along with parent-child and sibling relational problems. For example, parental depression and substance use can lead to serious detrimental effects on the quality of the parent-child relationship and contribute to broader family dysfunction; indeed, parents with depression and substance abuse have been found to be at greater risk for parent-child relational problems (Bailey et al., 2013; Beach et al., 2006b; Cummings, DeArth-Pendley, Du Rocher Schudlich, & Smith, 2001; Goodman & Gotlib, 1999). Parental antisocial behavior is also known to cause disruption in the parent-child relationship (Dutton, Denny-Keys, Sells, 2011; Wilson & Durbin, 2012).

Child Maltreatment Child maltreatment, including physical abuse, sexual abuse, neglect, and/or psychological abuse, may be present alongside any relational disorder. If child maltreatment is also present, Slep and Tamminen (2012) recommend that it be labeled as the primary relational category as the responsibility for appropriate treatment of children lies with the caregiver. The authors argue that labeling child maltreatment as a parent-child relational problem could negatively affect treatment decisions and inappropriately shift responsibility away from parents to both child and parent.

In summary, numerous disorders and conditions have been associated with parent-child and sibling relational disorders. Clinicians ought to assess carefully for a range of disorders in both children and parents, when relational problems are present.

Course and Developmental Challenges

As described above, parent-child and sibling relational problems often co-occur with individual diagnoses and impairments, and thus the course of relational problems often depends in part on the successful treatment (or lack theoreof) of the individual diagnoses. In addition, distinguishing between suboptimal, dysfunctional parenting that is common in parent-child and sibling relational problems, and child maltreatment, can pose challenges (Wolfe & McIsaac, 2011). While more severe forms of parenting problems such as child maltreatment have known negative effects for child development that are well documented (e.g., Cicchetti & Toth, 2005; Kaplow & Widom, 2007; Pollak, 2008; Tarullo & Gunnar, 2006; Watts-English, Fortson, Gibler, Hooper, & DeBellis, 2006), less has been established regard-

ing the developmental consequences of milder forms of family dysfunction, as more likely captured within the parent-child and sibling relational problem categories. Still, the early family environment is known to have significant impact on the development of the child, particularly with regard to brain development and neuroendocrine stress reactions (Fox, Levitt, & Nelson, 2010; Gunnar, 1998), with later consequences for attachment, stress reactivity, and emotion regulation (Gunnar, Brodersen, Nachmias, Buss, & Rigatuso, 1996).

In addition, there is evidence that coercive family cycles that start with the parent-child and sibling relationships often generalize to the peer realm, resulting in association with delinquent peers or social rejection by peers in later childhood and adolescence (Patterson, Forgatch, & DeGarmo, 2010; Patterson et al., 1991; Updegraff, Thayer, Whiteman, Denning, & McHale, 2005). For example, negative parenting can result in the child developing overt forms of antisocial behavior, such as excessive noncompliance and arguing. The behaviors learned in the home often generalize to social settings in the school and community. When a child displays noncompliance with teachers, academic failure is a likely outcome. Similarly, when children engage in such behavior with peers, children are rejected by their normative peer group. Such failures can lead to depressed mood and lead youth to affiliate with a deviant peer group whereby new forms of antisocial behavior are learned, such as stealing, drug use, or gang involvement (Dishion, Veronneau, & Myers, 2010; Patterson et al., 2010). In females, teen pregnancy and involvement in the child welfare system can result (Helfrich & McWey, 2013; Leve, Kerr, & Harold, 2013). In young adulthood, individuals who experienced parent-child or sibling relational problems early in life often fail to attain stable employment and/or have lower standards of living as indexed by their income or occupational status (Patterson et al., 2010).

Researchers are increasingly turning their attention to the mediating and moderating factors that increase the risk of the development of child psychopathology when parent-child or sibling relational problems exist (e.g., Gunnar, 1998; Laught, Esser, & Smidt, 2001). Wamboldt and Reiss (2006) delineate the role of relational processes for mediating and moderating genetic influences for the development of psychopathology in children, highlighting the importance of parent-child relationships within the interplay of gene-by-environment factors for risk and resilience. Particularly salient is evidence for the development of antisocial behaviors and conduct problems in adolescents. Through gene-by-environment interactions, parent-child conflict and harsh parenting may exacerbate an adolescent's underlying genetic risk for conduct problems (such as aggression or lack of impulse control) and contribute to the expression of antisocial behaviors (O'Connor, Hetherington, Reiss, & Plomin, 1995; Reiss, Neiderhiser, Hetherington, & Plomin, 2000). For example, a study by Caspi and colleagues indicated that the effects of maltreatment on antisocial behavior were stronger among males with a genotype conferring lower levels of MAOA expression (Caspi et al., 2002). Conversely, inherited sociability may serve to buffer a child from developing problems, even in the face of an insensitive, unresponsive caregiving environment (Van Ryzin et al., 2015). In addition, inherited qualities in the child can directly influence parenting and the parent-child relationship. This occurs when genetically influenced child characteristics evoke a response from their parents. Three separate adoption studies have demonstrated support for this evoked effect; two found that children whose biological parents had more antisocial behavior were more likely to receive negative or hostile parenting from their adoptive parents than children without genetic risk (Ge et al., 1996; O'Connor, Deater-Deckard, Fulker, Rutter, & Plomin, 1998), and a third found that children whose biological parents had more ADHD symptoms were more likely to receive more hostility from their parent (Harold et al., 2013). Similarly, twin studies have demonstrated a genetically evoked effect on parenting during infancy, childhood, and adolescence (Deater-Deckard, 2000; Deater-Deckard & Petrill, 2004; Forget-Dubois et al., 2007; Klahr, Thomas, Hopwood, Klump, & Burt, 2013;

McGue, Elkins, Walden, & Iacono, 2005; Reiss et al., 1995). These genetically informed studies suggest an intricate interplay between genes of the parent and child and the etiology and developmental course of parent-child relational problems.

Treatment and Treatment Outcomes

A number of evidence-based interventions that target parenting behaviors and address family ecological factors have been shown to effectively improve parent-child relationships and other child and adolescent outcomes. The following is a succinct review of several evidence-based interventions that may be relevant for the treatment of parent-child and sibling relational problems. Our review is not intended to be all-encompassing; rather, we highlight some of the more commonly implemented evidence-based interventions.

Parent-Child Interaction Therapy Drawing on both attachment and social learning principles, parent-child interaction therapy (PCIT) is an evidence-based treatment aimed at improving the parent-child relationship and decreasing disruptive behavioral problems in children and adolescents, such as oppositional defiant disorder and conduct disorder (Eyberg, Boggs, & Algina, 1995). The intervention involves actively coaching parents in relationship skills, authoritative parenting, and behavioral change skills; encouraging the development of a warm, nurturing relationship; and effective limit-setting and discipline practices (Zisser & Eyberg, 2010). Randomized control trials have demonstrated the effectiveness of PCIT, indicating fewer child behavioral concerns and increases in child compliance, positive parenting, and positive parent-child interactions as a result of treatment (Bagner & Eyberg, 2007; Schuhmann, Foote, Eyberg, Boggs & Algina, 1998). Follow-up studies have found maintenance of these treatment effects over time (Boggs et al., 2005; Eyberg et al., 2001; Hood & Eyberg, 2003). PCIT also has demonstrated effectiveness as treatment for physically abusive parents (Chaffin et al., 2004), with those families receiving PCIT showing fewer coercive parent-child interactions and fewer reports of child abuse after treatment than families in the control condition.

Parent Management Training—Oregon Model (PMTO) Informed by a social interaction learning model, the Parent Management Training—Oregon Model (PMTO) includes multiple interventions for the prevention and treatment of antisocial behavior in children and adolescents. Developed in the early 1970s, scientists at the Oregon Social Learning Center have overseen the continual evolution and evaluation of the PMTO model (Forgatch & Patterson, 2010). Connecting antisocial behaviors in children to the influence of the family environment, the interventions target patterns of parent-child interactions in order to decrease child and parent coercion and increase positive parenting behaviors (Forgatch & Patterson, 2010). Such positive parenting behaviors include limit-setting, monitoring, skill encouragement, problem-solving, and positive involvement. Through highly structured yet flexible sessions, therapists work with individual families to develop and practice new skills, tailoring content to each family's needs. The effectiveness of PMTO has been well established through numerous randomized control trials (see Forgatch & Patterson, 2010), demonstrating a decrease in antisocial and deviant behaviors in children and adolescents and an increase in positive youth outcomes.

EcoFIT and the Family Check-Up A developmentally informed ecological intervention, the multilevel EcoFIT model aims to improve child and adolescent outcomes and decrease risk factors for problem behaviors by intervening at the school and family levels (Dishion & Stormshak, 2007). The Family Check-Up (FCU) is an evidence-based family-level component of EcoFIT aimed at increasing effective parenting and family management practices and decreasing problem child and adolescent problem behavior and substance use (Stormshak & Dishion, 2009). Using a strengths-based approach, the FCU provides a comprehensive, ecological assessment of family and child functioning and uses motiva-

tional techniques to address parent readiness for change. Depending on the unique needs of each family, the intervention offers family management training that targets the development of key parenting skills such as positive reinforcement, effective limit-setting, monitoring, communication, and problem-solving. Twenty-five years of research, including numerous randomized control trials, have demonstrated the effectiveness of the FCU for improving family management practices and reducing risky and problem behaviors in children and adolescents, including substance use (Dishion, Kavanagh, Schneiger, Nelson, & Kaufman, 2002), early childhood conduct problems (Shaw, Dishion, Supplee, Gardner, & Arnds, 2006), and child mental health concerns such as depression (Connell & Dishion, 2008).

The Incredible Years Training Series The Incredible Years Training Series (Webster-Stratton, 1996; Webster-Stratton & Reid, 2003), a parent training program that targets parents with children ages 0–13 years, focuses on the increase of positive parenting behaviors and the decrease in negative parenting behaviors. The training includes parents, teachers, and children and is intended as an early intervention to prevent and treat child problem behaviors at onset (Webster-Stratton & Reid, 2010). Through the use of videotapes and therapist interaction, parents are trained to increase parenting competency, decrease the use of harsh discipline, and learn positive communication and problem-solving skills to use with their children. A number of randomized control trials have demonstrated the efficacy of The Incredible Years for decreasing child behavioral problems, reducing the use of harsh discipline and coercive parenting, and improving parent-child interactions and bonding (Webster-Stratton & Reid, 2010).

Functional Family Therapy Functional Family Therapy (FFT), an evidence-based family systems and behavioral intervention, has been widely disseminated and is aimed at treating families of youth with disruptive behaviors such as conduct disorder and delinquency, as well as substance use disorders (Alexander et al., 1998; Waldron, Slesnick, Brody, Turner, & Peterson, 2001). The ecological model underlying FFT posits that substance use and behavioral problems in adolescents are developed and maintained within maladaptive family environments (Alexander et al., 1998). Over the course of 12–16 sessions, the intervention targets family interactions and relationships and uses behavioral and cognitive strategies to enhance the family ecology and decrease problem behaviors (Waldron & Brody, 2010). Outcome studies, including numerous randomized control trials, have demonstrated the effectiveness of FFT for decreasing substance use as well as a wide range of problem behaviors among adolescents (Alexander et al., 1998).

Sibling-Based Interventions Recently, a number of interventions have been developed that have a specific focus on the sibling relationship and apply the same principles as discussed above in the PMTO intervention model. One such intervention, "Siblings are Special," is aimed at preventing behavior problems by enhancing youths' social and emotional competencies in the context of the sibling relationship, as well as enhancing parents' ability to manage sibling relationships (Feinberg et al., 2013). Using an after-school format for the siblings and family night sessions for families, this intervention resulted in enhancing positive sibling relationships and appropriate strategies for parenting siblings. A second intervention aimed specifically at siblings in foster care has also shown positive effects on the sibling relationship (McBeath et al., 2014). In addition to interventions that directly target the sibling relationship, some family-based interventions that have not had a specific focus on the sibling relationship also been shown to have a spillover effect onto the sibling relationship. For example, a family-based intervention aimed at preventing conduct problems in preschoolers showed positive effects in reducing antisocial behavior among the preschoolers' adolescent adjudicated sibling (Brotman et al., 2005).

The aforementioned interventions are among the most commonly employed evidence-based programs for parent-child and sibling relational problems that have not reached the level

of a clinical diagnosis. Other evidence-based interventions that more specifically target clinical problems may be appropriate for the treatment of relational problems, particularly when comorbid diagnoses are present. Other widely implemented evidence-based programs for clinical-level problems include Multisystemic Therapy (see Henggeler & Schaeffer, 2010), Trauma-Focused Cognitive Behavioral Therapy (TF-CBT; see Cohen, Mannarino, & Deblinger, 2010), Attachment-Based Treatment (Dozier, 2003), Multidimensional Treatment Foster Care (Chamberlain, 2003), and Brief Strategic Family Therapy (see Robbins, Horigan, Szapocznik, & Ucha, 2010), to name a few.

Summary

Parent-child and sibling relational problems can be serious issues that impact the mental health of the child and/or parent and are impacted by the mental health of each. Although neither relational disorder is considered a diagnosis in the *DSM-5*, there has been increasing attention to relational problems with each iteration of the *DSM*. Further, it is likely that relational problems co-occur with one or more diagnoses in either or both of the individuals in the relationship. Overall, the prevention and intervention work described in this chapter has suggested that, with early and appropriate assessment and intervention, parent-child relational problems can see marked improvement over time. Nonetheless, because of the high likelihood of comorbid disorders in the parent and/or child when parent-child relational problems are present, it is important for clinicians to attend to other diagnoses and to treat them in accordance with the recommendations outlined in other chapters in this volume. At times, pharmaceutical interventions may be warranted to help treat an underlying disorder, in conjunction with the psychosocial interventions detailed above. Without treatment, there is an increased risk that relational problems will lead to intergenerational continuity of problems into the next generation (Raudino, Fergusson, Woodward, & Horwood, 2013).

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Family Disruption

30

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General Overview of Family Disruption

Decades of research have documented that family disruption, such as family separation, divorce, and marital discord, can have detrimental effects on children's mental health and well-being (Lee & Gotlib, 1991; Rutter, 1984). More recently, other forms of family disruption have been a source of inquiry, particularly with respect to parent deployment and veteran suicide and resulting impacts on children's functioning (Gorman, Eide, & Hisle-Gorman, 2010; Manos, 2010). Importantly, children who experience family disruption are at higher risk of a variety of mental health difficulties, including depression (Gilman, Kawachi, Fitzmaurice, & Buka, 2003; Lamb, 1999), anxiety (Slater, Stewart, & Linn, 1983; van Gils, Janssens, & Rosmalen, 2014), as well as the potential for academic failure (Somers et al., 2011). Given the rate of divorce in the United States hovering around 50% (Forste & Heaton, 2004), the number of children in the child welfare system, and the proportion of children living in poverty, the implications on a myriad of child outcomes are of paramount concern (Lamb, 1999).

With the advent of the *Diagnostic and Statistical Manual of Mental Disorders, 5th Edition* (American Psychiatric Association, 2013), the means by which family disruption was categorized among mental health researchers and professionals changed. Specifically, some forms of family disruption are now categorized into V-codes. This represents a change from the 4th edition of the Diagnostic Manual (APA, 2000) in that the multiaxial system was removed, and therefore the family disruption categories that were once represented on Axis IV in Psychosocial and Environmental Problems were by and large moved into available V-codes. For instance, "problems with primary support group" was an Axis IV category in which death of a family member, disruption of a family by divorce or separation, or child removal from the home would have typically been represented, particularly given how these circumstances may contribute to, exacerbate, or maintain mental health diagnoses. Existing V-codes in DSM-V that map onto these categories are primarily represented in the *relational problems* section of

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"other conditions that may be a focus of clinical attention." These include *upbringing away from parents (V.61.8)*, *disruption of family by separation or divorce (V61.03)*, *uncomplicated bereavement (V62.82)*, and *child affected by parental relationship distress (V61.29).* In addition to these V-codes, however, are other forms of family disruption that may not be clearly categorized, such as disruption by parent deployment and living with a parent with mental illness, among others.

Given the importance of key relationships in children's lives, the impacts of threats to these relationships can have significant detrimental effects. Thus, detailing the variations in relationship threats in any form is likely to be highly relevant to the expression of children's mental health difficulties. However, the relationship between family disruption and deleterious mental health outcomes can vary and may be mediated by a number of factors. These factors may act as either risk or protective variables in the expression of children's mental health difficulties, as well as have implications for prognosis should a mental health condition be diagnosed. For example, a general, prevailing theory that family disruption of any kind may affect parental availability and/or responsivity to a child is considered a cross-cutting, principal risk factor that increases the potential for negative outcomes (Lee & Gotlib, 1991).

Indeed, the effect of family disruption is significant, given the tendency for disruption to heighten parents' self-focus and reduce abilities to provide appropriate supervision and nurturing for their children. These impacts may even form the basis for intergenerational consequences of family disruption (McLanahan & Bumpass, 1988). In contrast, other research examining mediating factors of family disruption has found that the presence and support of a father may buffer children's mental health difficulties, even above and beyond the impacts of children's involvement in the child welfare system (Somers et al., 2011). Given these variations and the possibility of important protective factors that could be a direct focus of public policy and mental health prevention efforts, understanding the defining features, nuances, and empirical research of each major type of family disruption is warranted.

Upbringing Away from Parents

DSM Designation

The DSM-V criterion for the designation "upbringing away from parents" is considered a *relational problem* and can be found in the DSM section, "other conditions that may be a focus of clinical attention" (APA, 2013). It is described as a:

> category that should be used when the main focus of clinical attention pertains to issues regarding a child being raised away from the parents or when this separate upbringing affects the course, prognosis, or treatment of a mental or other medical disorder. This could be one who is living in a non-parental relative's home, or with friends, but whose out-of-home placement is not mandated or sanctioned by the courts. Problems related to a child living in a group home or orphanage are also included. This category excludes issues related to V60.6 (Z59.3) children in boarding schools. (p. XX)

Currently, the number of children in the foster care system hovers around 400,000, with approximately half of these children represented by the male gender and racial minorities (US Department of Health and Human Services Administration for Children and Families, 2014). Adolescents are more likely to be placed in foster care than are infants or school-age children (40% adolescent entry rate) (Wulczyn, Hislop, & Harden, 2002), although children under the age of 5 are most likely to suffer neglect and maltreatment compared to other age groups, thus placing this age group at the most risk of being removed due to these issues (Pinderhughes, Harden, & Guyer, 2007). There are multiple forms of foster care placements, including nonrelative care, kinship care, group homes, or residential institutions, as well as informal placements that are not documented by state or federal agencies. However, children living in educational boarding schools are excluded from the same DSM designation.

The "upbringing away from parents" designation was added to the DSM-V in order to differentiate forms of relational issues that may be present within caregiving relationships.

Emotional and Behavioral Outcomes

A large proportion of youth who are removed from their parents' homes struggle with emotional or behavioral problems. However, it varies as to whether or not these difficulties were present prior to removal or developed as a result of the removal (Hussey, Falletta, & Eng, 2012). Given that neglect and maltreatment is a predictor for youth being removed from their parents' homes, a subset of these individuals enter into the child welfare system with existing emotional or behavioral problems resulting from their abuse experiences, such as posttraumatic stress symptoms or problems with anxiety (Hussey et al., 2012). At the same time, research has demonstrated that a child's upbringing away from their biological parents may be a cause for developing mental health and behavioral problems (McCrae, 2009). As described by Bronfenbrenner and Morris (2006), development is affected in dynamic ways, whereby the individual child has characteristics that impact their environment (e.g., demographic characteristics, genetic predispositions) and the environment, in turn, impacts an individual child. This interactional process occurs through multiple levels of a child's ecological system; however, interactions at the microsystem, in which family members exist, are most frequent and may be most impactful (2006; Pinderhughes et al., 2007). Due to these complex systems, the ability to determine if psychopathology developed before or after a child experiences upbringing away from their parents is a bit of a "chicken or the egg" determination for any group of young people. In addition, youth who enter the foster care system are additionally at risk of being further abused within foster homes, thus contributing to additional developmental insults (Pinderhughes et al., 2007).

In general, what *is* clear is that youth who meet the DSM-V designation indicating that there has been disruption in their living environments are vulnerable to specific externalizing and internalizing difficulties. Indeed, studies have consistently demonstrated that disruptive behavior disorders are overrepresented among foster care youth, even in comparison to other pediatric clinical populations (Dore, 1999; Pilowsky, 1995). Though disruptive behavior problems are defined in varying ways in the literature, including utilizing different assessment and identification methods, the finding that externalizing problems are the most common form of psychopathology among youth within the child welfare system is well accepted (Keil & Price, 2006). This is particularly concerning given that youth with externalizing problems are likely to remain in the child welfare system and be separated from their family of origin for longer periods of time than youth without these difficulties (Keil & Price, 2006). Prevalence rates vary by study, however, with estimates ranging anywhere from 20% to nearly 80% of youth in child welfare having diagnosable disruptive behavior problems (Keil & Price, 2006; Silver et al., 1992; Thompson & Fuhr, 1992). The severity of the externalizing difficulties also often varies by type of out-of-home placement. For example, Helfinger, Simpkins, and Combs-Orme (2000) found that youth in residential placements tended to have more severe conduct problems, whereas youth in other settings such as foster care or kinship care had less severe conduct problems or other externalizing difficulties such as oppositional defiant disorder (ODD) or attention deficit-hyperactivity disorder (ADHD). In addition, although racial minorities are disproportionately present in the child welfare system, certain minority groups may be more likely to receive services as a result of their externalizing difficulties. In a study by Gudino, African American youth were more likely to receive services than non-Hispanic white youth when externalizing problems were in their diagnostic profiles (2012).

Treatment and Recommendations

Despite the degree of risk among children who are raised outside of their parents' homes, recent

research has identified a number of caregiver characteristics and environmental factors that can promote resilience. First and foremost, the systems in which children are involved when they are raised outside of their parents' homes are often fragmented, and the risk for these youth to be subjected to unstable placements, abusive foster care situations, and uncertain futures is high. Ungar, Liebenberg, and Ikeda (2014) described these aspects of the child welfare system via a review of case records ($N = 116$) and provided a description of "typical" youth within the system. From this work, the authors were able to make recommendations based on their case reviews, interviews, and file reviews with multiple providers involved in each case. Systemic insufficiencies, combined with disproportionate service utilization, lead to specific themes regarding methods for improving the long-term outcomes for youth and families. For instance, developing interventions across systems for children being raised away from their parents need to be multilevel to address variations in need complexity, coordinated across interventions and organizations (i.e., teachers, mental health providers, and primary caregivers), provide continuity of care, and be stakeholder-informed.

In addition to system-level recommendations, a recent qualitative study by Storer, Barkan, Stenhouse, Eichenlaub, Mallillin, and Haggerty (2014) assessed children, child welfare staff, and foster caregivers for the strengths and challenges associated with child-caregiver relationships and providing temporary homes for children in the system. Their results showed that the most successful homes fostered a sense of belongingness and inclusiveness for youth, coupled with structure and consistency in parenting techniques. Other research has demonstrated that characteristics of the caregiver, such as those described above, individual youth characteristics (e.g., intelligence, curiosity, pleasing appearance; (Radke-Yarrow & Sherman, 1990), as well as a child's positive relationship with their child welfare worker statistically predicted resilience in multiple child domains including behavioral and academic functioning (Bell, Romano, & Flynn, 2013). Taken together, these studies suggest that by bolstering adults, in particular, functioning within the child welfare system (caregivers, staff/workers) combined with improvements in coordinated care, youth can maintain or bolster their resilience to the deleterious effects of being raised away from their biological parents.

Disruption of Family by Separation or Divorce

Prevalence

Multiple estimates of family constellation suggest that half of all marriages will end in divorce, which is in sharp contrast to data a century ago in which less than 5% of marriages ended in this manner (Forste & Heaton, 2004). There are multiple consequences of divorce, particularly for marriages that end in which children are involved. Not only is the mental health of children subject to divorce at risk, but they may also suffer from social (e.g., changing schools) or physical consequences (e.g., change in healthcare providers), as well as long-term problems related to socioeconomic status (Forste & Heaton, 2004). A number of researchers have posited theoretical perspectives on the pervasive impact of divorce on children, with the most well supported being those that address the complexity within family systems, such as the Minuchin Family Stress Model (Pardeck, 1989). The model proposes that families are ecological units in which our understanding of risk and resilience factors is dependent upon the dynamic, interactive aspects of the systems. Although there are individual factors present, theories such as the Minuchin Family Stress Model emphasize that those factors do not exist in a vacuum – rather they affect each level of an ecological unit which, in turn, impacts any one individual. Thus, when disruption occurs in spousal relationships, the resulting stresses impact children in the distressed families as well. In turn, children's responses to these stresses may negatively impact parents, and these interactional effects may predict adjustment problems or long-term difficulties as a result.

DSM Designation

Regardless of the theories posited to predict adjustment difficulties following divorce, the empirical literature in children's mental health generally suggests that a myriad of factors are related to negative divorce outcomes for youth, necessitating a DSM V-code distinction. As with the "upbringing away from parents" distinction, "disruption of family by separation or divorce" was moved into the *other conditions that may be a focus of clinical attention* listed as a V-code when the axial system was dropped in DSM-V (APA, 2013).

Emotional and Behavioral Outcomes

Some research indicates that children's peri- or postdivorce adjustment may be predictive of preexisting mental health states, rather than the divorce itself, and that some behavioral or emotional difficulties may resolve as a result of divorce (Wadsby & Svedin, 1993). However, a large body of literature has focused primarily on children's poor adjustment to divorce, given the likelihood that significant distress is associated with separation and divorce. For example, a peer-reviewed journal, the *Journal of Divorce and Remarriage*, was developed specifically to detail some of these issues, as well as the adult-specific factors, and has published more than 50 issues.

Because the child outcomes related to divorce are complex and may vary from study to study, researchers have attempted to understand the varying relationships between individual and ecological factors that may predict positive or negative adjustment. For instance, a recent study by Weaver and Schofield (2014) utilized data from the National Institute of Child Health and Human Development Study of Early Child Care and Youth Development in order to assess the mediating and moderating factors associated with children's emotional and behavioral problems related to divorce. Their findings revealed that divorce is associated with significantly more externalizing behavioral problems in children of divorced families than in children from intact families. Children of divorce also struggled with internalizing difficulties, such as anxiety and depression; however, socioeconomic status moderated this relationship in that children from families with higher socioeconomic status prior to divorce had fewer internalizing difficulties overall. The researchers also assessed mediators and determined that maternal depression, low environmental support and stimulation, and lower socioeconomic status predicted higher levels of postdivorce externalizing problems. Importantly, when children had higher IQ's and their mothers exhibited higher levels of sensitivity and nurturance, these were generally considered protective factors. In sum, this research suggests that certain individual factors may predict coping abilities related to divorce, though external factors such as parental characteristics and parents' ability to create a supportive, nurturing environment postdivorce may both explain many children's adjustment difficulties as well as pinpoint areas for prevention efforts.

Treatment and Recommendations

Though there are treatments for children struggling with their parents' divorce, therapies with the most evidence are primarily targeting the emotional or behavioral problems manifested by the divorce and subsequent environmental circumstances. For instance, if children are struggling with internalizing symptoms, such as anxiety or depression, front-line treatments for those difficulties would be most appropriate. Divorce situations could be used in those therapy contexts to build skills relevant to copying with anxiety- or depression-related symptoms. Given that externalizing difficulties are correlated with both pre- and postdivorce functioning, a number of available treatments share therapy modalities with emphasis on parent management training skills, increasing environmental consistency, addressing parental availability, and individual skill-building related to relaxation training and anger management (Bornstein, Bornstein, & Walters, 1988; Pedro-Carroll & Cowen, 1985; Stolberg & Walsh, 1988; Stolberg & Mahler,

1994). Researchers also recommend policies that support the financial and emotional well-being of the residential parent to enhance child outcomes (Lamb, 1999). By addressing the environmental influences on children's adjustment to divorce, while bolstering their individual coping abilities, treatment programs can be effective at improving the mental health of children experiencing divorce as a form of family disruption. Another "chicken or the egg" question related to family disruption is whether divorce itself is the cause for adverse child outcomes or, alternatively, if the marital conflict leading up to the divorce is the cause for these outcomes.

Child Affected by Parental Relationship Distress

DSM Designation

Though not entirely distinguishable from the literature on children affected by divorce or separation, the fifth edition of the DSM also includes a category for youth impacted by parental relationship distress (APA, 2013). Specifically, "this category should be used when the focus of clinical attention is the negative effects of parental relationship discord (e.g., high levels of conflict, distress, or disparagement) on a child in the family, including effects on the child's mental or other medical disorders" (p. XX). As with the other categories focused on children and found within the *other conditions that may be a focus of clinical attention* section, the primary reason a clinician may specify the c*hild affected by parental relationship distress* V-code is in relation to its interaction or effects with existing or correlated mental health or medical problems.

Emotional and Behavioral Outcomes

Parent relationship distress is associated with a number of negative mental health sequelae for children. Psychopathology that may occur in childhood, such as anxiety disorders, disruptive behavior disorders, and depression, have all been correlated with parent relationship distress (Dadds & Powell, 1991; Gerard, Krishnakumar, & Buehler, 2006). However, individuals suffering from longer-term difficulties starting in adolescence or adulthood, such as substance abuse, criminal involvement, and borderline personality disorder, are also more likely to have been subject to their parents' relationship distress in their childhood, based on retrospective reports (Mandelbaum, 1980; Sanders & Ralph, 2005). Not surprisingly, parent relationship distress is highly correlated with divorce; thus, the resulting difficulties relevant to the impact of divorce on children are also relevant to relationship distress in general. As with multiple areas of family disruption, when parents' relationship is a source of stress for one or both caregivers, their ability to allocate mental and physical resources toward parenting suffers and may result in inconsistency, poor supervision, as well as individual caregiver mental health problems associated with childhood behavioral or emotional problems, such as depression.

Treatment and Recommendations

Though few treatments exist specifically for children experiencing parental relationship distress, the fact that the difficulties associated with this category are thematically similar to divorce suggests that similar treatment approaches would be equally effective. In addition, barring relationship/couple's counseling, the emphasis in treatments for youth experiencing these difficulties should be focused on how children's problems related to their parents' relationship problems manifest. Thus, if externalizing behavioral issues are primary, treatment should be focused on parent management training in light of household inconsistencies, reduced structure, etc. (Dwairy, 2010). In addition to distress related to separation from family members due to family conflict, children are also likely to experience distress related to the loss of family members and loved ones resulting from death as described in the section below.

Uncomplicated Bereavement

DSM Designation

Bereavement describes the state of individuals who have lost a loved one due to death and was first included in the *Diagnostic and Statistical Manual of Mental Disorders, 3rd Edition* (DSM-3 (American Psychiatric Association, 1981)) as an exclusionary criterion after a series of studies suggested that, while symptoms of depression were common during bereavement, symptom presentation was often brief and not requiring treatment and therefore not suitable for a depressive disorder diagnosis (Zisook et al., 2012). Therefore, a "bereavement exclusion" criterion was included in the 1980 publication of DSM-3 dictating that a major depressive episode occurring less than 2 months following the death of a loved one would not be diagnosed if symptoms could be better accounted for by bereavement (APA, 1981). However, over the last 30 years, research has accrued suggesting that bereavement-related depression is similar to major depression in presentation, patterns of comorbidity, and risk factors associated with onset and chronicity such as genetic influence (APA, 2013). Therefore, the bereavement exclusion criterion was removed from the *Diagnostic and Statistical Manual of Mental Disorders, 5th Edition* (DSM-5) (APA, 2013) and bereavement can now be diagnosed as part of a major depressive episode or as V62.82 (Z63.4) *uncomplicated bereavement* which involves:

> …a normal reaction to the death of a loved one. As part of their reaction to such a loss, some grieving individuals present with symptoms characteristic of a major depressive episode—for example, feelings of sadness and associated symptoms such as insomnia, poor appetite, and weight loss. The bereaved individual typically regards the depressed mood as "normal," although the individual may seek professional help for relief of associated symptoms such as insomnia or anorexia. The duration and expression of "normal" bereavement vary considerably among different cultural groups. (APA, 2013, p.XX)

Additionally, if symptom presentation exceeds normal bereavement, occurs within 3 months of the death, and abates within 6 months of the death, an adjustment disorder many be diagnosed. However, if symptoms are clearly a response to bereavement but presentation does not align with adjustment disorder (e.g., symptom onset occurs more than 3 months after the stressor or continues for more than 6 months following the stressor), then other specified trauma- and stressor-related disorder may be diagnosed with a persistent complex bereavement disorder specifier (APA, 2013).

Emotional and Behavioral Outcomes

Symptom presentation among bereaved individuals varies. When a traumatic event produces a violent and unexpected death, symptoms of post-traumatic stress disorder or acute stress disorder may be present. Symptom presentation following bereavement may also align with depressive disorder presentations; changes in appetite, sleep pattern, and weight; and low mood (APA, 2013). Importantly, however, DSM-V distinguishes grief from a major depressive episode based on the predominating affect. Specifically, a major depressive episode involves persistent depressed mood and inability to anticipate happiness or pleasure. In contrast, grief presents with feelings of emptiness and loss as well as dysphoria that is likely to decrease in intensity within days or weeks but can also be accompanied by positive emotions and humor. However, similar to non-bereaved depression, the risk of mortality among persons experiencing bereaved depression is higher compared to non-bereaved individuals matched for age and gender (Stroebe & Stroebe, 1993). Therefore, symptoms associated with mortality risk should be assessed and monitored throughout the course of treatment.

Treatment and Recommendations

Because uncomplicated bereavement and depression due to bereavement were only recently designated diagnosable disorders, there is currently little available literature involving the new diagnoses. However, the impact of

bereavement and grief on family disruption has been explored in detail (Stroebe, Hansson, Stroebe, & Schut, 2001). Importantly, experts in the field of bereavement and grief continually conclude that routine psychotherapeutic or pharmacological intervention for bereaved individuals is not a given (Raphael, Minkov, & Dobson, 2001). Indeed, there is currently no empirical evidence supporting routine or obligatory intervention simply because someone has experienced bereavement (Stroebe, Folkman, Hansson, & Schut, 2006)

Bereavement treatment may occur at the individual, couple, and/or family system level. Research suggests that often the most enduring and stressful bereavement experience occurs in parents who survive the death of a child (Rubin & Malkinson, 2001; Shimshon Rubin, 1999). The severity of parental bereavement centralizes on three thematic issues: (1) the loss of a person who was part of the family system, (2) the loss of a sense of personal power and competence, and (3) the loss of a part of the self (Malkinson & Brask-Rustad, 2013). Therefore, cognitive behavior couple's therapy, particularly as it relates to the death of a child, focuses on normalizing the grief process as well as the unique nature of individuals' grieving progression, promoting and enhancing couple's communication skills, and easing distressing symptoms (Malkinson & Brask-Rustad, 2013).

Similar to couple's treatment, treatment for bereaved children typically entails normalizing the child's experience of bereavement, developing coping mechanisms for changes to the child's environment including family and peer dynamics, encouraging the child to communicate concerns or troubles, and discussing and clarifying death-related issues that might seem confusing or frightening (Lohnes & Kalter, 1994). Other treatment modalities have been developed, such as art therapy (Lister, Pushkar, & Connolly, 2008), play therapy (McMahon, 2009), and music therapy (Mondanaro, 2005), which assist the child through the grieving process though the evidence base for these treatments are less established.

Prognosis and Outcome

Research suggests that, as applied to the bereaved, mental health prognosis and outcome is best predicted by assessing multiple risk and protective factors both proceeding and following the death (Sandler, Wolchik, & Ayers, 2008; Stroebe et al., 2001). Notable protective factors for parentally bereaved children include quality parenting by the surviving caregiver (Raveis, Siegel, & Karus, 1999), absence of additional stressful events (Thompson, Kaslow, Price, Williams, & Kingree, 1998), and caregiver mental health (Lin, Sandler, Ayers, Wolchik, & Luecken, 2004). In a systematic review of bereavement health outcomes, researchers found that compared to non-bereaved counterparts, bereaved individuals are at a greater risk for mortality from many causes, including suicide; have higher rates of disability, hospitalization, and medication use; and are at a greater risk for developing psychiatric conditions, especially in early bereavement (Stroebe, Schut, & Stroebe, 2007). Importantly, however, the estimated incidence of prolonged intense grief in the general population is only 11–15% (Macias et al., 2004).

Other Forms of Disruption

Deployment Stress

Children and families experience a myriad of other forms of family disruption that warrant clinical attention but are not included as formal DSM diagnoses. With recent conflict in the Middle East, millions of children and families are experiencing the stress of family member deployment (McFarlane, 2009). In DSM-V, *problem related to current military deployment status* (V62.21) is used when an occupational problem directly related to an individual's military deployment status is the focus of clinical attention or has an impact on the individual's diagnosis, treatment, or prognosis. Psychological reactions to deployment are not included in this category; such reactions would be better captured as an

adjustment disorder or another mental disorder (APA, 2013).

When applied to children and families of the deployed, diagnoses are formed from presenting symptoms and often include anxiety, depressive, or adjustment disorders. Unique stressors including moving every 2–3 years; media coverage of wartime events, particularly during a family member's deployment; and adapting to the absence and later re-establishment of the deployed member in and out of the family system are only some of the many stressors imposed upon military families (Huebner, Mancini, Wilcox, Grass, & Grass, 2007; McFarlane, 2009; US Department of Defense, 1998). A recent review of interventions for children and families experiencing family discord related to military deployment suggests that social support during deployment and reintegration of the military member is key to assisting with family relationship discord (McFarlane, 2009). Further, summer residential programs for children and family treatment approaches involving identity development, normalizing reactions to deployment and reintegration, improving interpersonal communication, and encouraging intimacy through emotional engagement are useful treatment strategies (McFarlane, 2009).

Parent Psychopathology

There are a number of DSM-V diagnoses involving the mental health effects of families, especially children, coping with parent psychopathology (APA, 2013). Appropriate diagnoses are based on the situational and environmental experiences and might include *parent-child relational problem* (V61.20), child maltreatment- and neglect-related diagnoses, or *relationship distress with spouse or intimate partner* (V61.10) (APA, 2013).

Treatment for children and families experiencing parent psychopathology vary and are largely based on the specific psychopathology and associated direct effects on the family. For example, there is an abundance of literature involving children living with a substance-abusing parent and the impact that family environment has on subsequent psychological health as an adult (Easley & Epstein, 1991; Eiden, Leonard, Hoyle, & Chavez, 2004). Risk factors associated with poor adult functioning following a childhood with an alcoholic parent include passive appraisal of the problem, self-blame, and family disruption (Easley & Epstein, 1991). Therefore, support groups for children and families of alcoholic parents, such as Al-Anon or Alateen, emphasize therapeutic factors such as psychoeducation and social support (Cierpiałkowska, 1994).

Children coping with the stress of parental depression display high rates of anxiety, depression, and aggression and respond to family stressors in ways that are associated with symptoms of psychopathology (Langrock, Compas, Keller, Merchant, & Copeland, 2002). Importantly, children in these situations are more psychologically healthy when engaging in positive thinking, acceptance, and distraction instead of rumination and intrusive thinking (Langrock et al., 2002). Therefore, monitoring child mood lability (Birmaher et al., 2013) and cognitive processes (Langrock et al., 2002) is essential to treatment.

Conclusion

The effects of family disruption on children's mental health are numerous and varied. As described in the current chapter, various DSM designations exist in the 5th edition in the form of V-codes, primarily, though no one diagnosis encapsulates the myriad impacts of disruption on youth (American Psychiatric Association, 2013).

Often these designations are utilized in diagnostic profiles to explicate the core theme(s) underlying psychopathology or to alert professionals as to the historical aspects of a child's life and development. Importantly, multiple resilience factors can prevent the development of mental health difficulties related to family disruption; thus, the existence of a disruption "type" in a child's history is not equivalent to a diagnosis nor may warrant to inclusion of a V-code designation.

A majority of the clinical research, however, focuses on the risk factors and negative sequelae relevant to being removed from one's home, being exposed to divorce or parental estrangement or conflict, grief, and loss, or other factors such as parent deployment. Many youth struggle with a cascade of events following and related to initial disruption, such as multiple foster care placements, breakdowns in parenting skills or consistency, exposure to parental conflict, emotional unavailability of surviving caregivers, as well as the potential for these disruptive experiences to overwhelm their ability to cope resulting in individual mental health difficulties.

Much of the treatment literature is focused on addressing the mental health symptoms resulting from disruption, rather than treatments directly targeting the disruption. In some cases, such as with bereavement, treatment may not be warranted in order to avoid interrupting a normal coping process or to prevent overpathologizing normal reactions to loss (Stroebe et al., 2006). Certain forms of family disruption, such as divorce, occur so commonly and the mental health outcomes are similar enough that specified treatments have been developed to be administered in common settings such as schools (Stolberg & Mahler, 1994). The treatment literature is weakest, however, with regard to the cumulative effects of multiple forms of family disruption (Masten & Wright, 1998). Though available evidence-based treatments for severe mental health outcomes that are highly correlated with multiple disruption factors are effective, they are more limited in number than those targeting individual problem areas (Henggeler, Melton, & Smith, 1992).

One theme that emerged from review of the family disruption literature is that youth outcomes are most positive when caregiver consistency and the structure that available caregivers can provide is high. Resilience is fostered in these circumstances, particularly when available caregivers are able to combine consistency and structure with praise and positive behavioral supports (Bell et al., 2013). Similar to how risk factors may compound, resilience factors can also be cumulative and may result in the best possible outcomes with regard to external circumstances in children's lives. Thus, professionals working with youth who have experienced family disruption should focus on implementing resilience-promoting structures, supports, and treatments within as many caregiving relationships as are available in order to foster the most positive outcomes possible.

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Part XIII

Treatment Approaches

Cognitive Behavioral Therapy

31

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Introduction

The release of the *Diagnostic Statistical Manual – Fifth Edition* (DSM-5) in May of 2013 has not been without controversy and commentary. In this latest edition, the diagnoses have been reorganized to reflect some of the scientific advances that have occurred over the past 13 years (American Psychiatric Association [APA], 2013). However, some critics argue that the lowered thresholds of diagnostic criteria changes will inappropriately increase the prevalence of diagnoses and pathologize certain populations in society (e.g., Strakowski & Frances, 2012). However, as we place these changes into perspective and look at the larger picture of how these individuals are treated, we see that the empirically based treatment models have little to nothing to do with the changes in the DSM-5.

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Complex internalizing and externalizing pathology can be especially difficult for both families and schools and can subsequently impact the child both academically and socially. Therefore, treating mental health professionals need to be especially skilled in treating these youngsters. In this chapter there will be several techniques discussed in treating both internalizing and externalizing disorders. While we will provide the reader with broad, basic cognitive behavioral techniques, there are also advanced treatment methods discussed to provide a spectrum of interventions.

Case Conceptualization

The first step to working with a child is to gather information that will help to generate and test hypothesis (Beck, 1995; Persons, 1989). Often the time spent on this process is less than ideal, but one way to help ensure this is emphasized is through developing a solid case conceptualization. The case conceptualization is a practical tool that allows therapists to tailor specific cognitive behavioral techniques to the individual child, which will positively impact treatment planning.

When conceptualizing difficult cases, it is important to embrace flexibility. Therefore, recognizing that the child may be psychologically complex and collecting as much data as possible to gain insight and hypotheses is a favorable approach to understanding the child.

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The treating mental health professional begins to look deeply into a new youth upon first meeting. This requires gathering ample information about why the youth has come in for treatment. The reasons the parent may provide may not always align with the true presenting problem. Therefore, developing a rapport with the youth in an attempt to define the presenting problem is primary. Persons (1989) has suggested to move from the general problem and break it down into more discrete problems, compartmentalizing them into their cognitive, behavioral, physiological, emotional, and interpersonal components.

Other major influences to consider in case conceptualization are the developmental milestones of the youth and any cultural variables that may affect symptom expression and treatment (Friedburg & McClure, 2002). Let's first delve into developmental milestones. Typical or atypical development of the child is important to case conceptualization as atypical development can place the youth at risk for both internalizing and externalizing behaviors. Similarly, a youth's ethnocultural background is significant to the case conceptualization, as minority status alone may represent a stressor (Carter, Sbrocco, & Carter, 1996; Tharp, 1991). There are secondary considerations to culturally diverse youth, such as poverty, oppression, marginalization, prejudice, and institutional racism that may affect these youth from non-majority cultures (Sanders, Merrell, & Cobb, 1999). These circumstances may contribute to cognitions and behaviors that are entrenched in the youth's problems.

Lastly, cognitive variables are clinically imperative to the case conceptualization. Cognitive variables are manifested in the way a youth explains or predicts an event; these are commonly called automatic thoughts. Automatic thoughts often serve as initial treatment targets as they are typically easily accessible and provide insight into the child's schemata (Friedburg & McClure, 2002). Schemata suggest the core organizing beliefs and personal meaning structures (Beck et al., 1979; Beck & Freeman, 1990). Understanding a child's schemata provides insight into the content of their cognitive processes, which allows the clinician to work on variables such as flexibility of automatic thoughts, interpersonal behavior, and responsiveness to treatment (Friedburg & McClure, 2002).

Cognitive Behavior Therapy with Internalizing Disorders

Anxiety Disorders

Anxiety disorders are common in children and adolescents, with prevalence estimated at 13%. This includes generalized anxiety disorder, separation anxiety disorder, and various phobias. Shared features of these disorders include chronic worry, escape and avoidance, negative self-appraisal, and physiological over-arousal. Various anxiety disorders are commonly comorbid in children (Wilmshurst, 2015; APA, 2013).

Graduated in vivo exposure is integral to the treatment of many anxiety disorders. Therefore, we will begin with a brief description of this technique as described by Hazlett-Stevens & Craske (2003). The rationale for exposure treatment is that repeated direct exposure to feared objects or situations reduces the conditioned fear response. At the start of treatment, the client identifies specific situations or objects that arouse anxiety. The therapist then explains the rationale for exposure and collaborates with the client to build a hierarchy of feared items, ranging from the least to the most fear-provoking. Exposure begins at the lowest level to increase the chance of success. During exposure, the client is asked to monitor the feelings of anxiety and to avoid even subtle escape mechanisms (e.g., averting the eyes). When the current level on the hierarchy no longer produces severe anxiety, the client is exposed to the next step on the hierarchy.

Generalized Anxiety Disorder

Children with generalized anxiety disorder (GAD) experience excessive worry around many aspects of their lives and are unable to control this worry (APA, 2013). The age of onset is usually between the ages of 8 and 10. In young

children, GAD is often comorbid with separation anxiety disorder, while in adolescence depression and specific phobia are often comorbid (Wilmshurst, 2015).

When treating very young children, it is recommended that the therapist first establish a working relationship with the parents. Parents may need empathy for their situation, advice for coping with guilt, and affirmations of hope for the future (Feinstein, Fielding, Udvari-Solner, & Joshi, 2009). Early sessions should focus on developing skills such as relaxation training to alleviate physical symptoms such as muscle tension and developing a hierarchy of anxiety-provoking situations (Keeton et al., 2013; Grover, Hughes, Bergman, & Kingery, 2006). When explaining the treatment rationale to children, the therapist should use stories and simple language to make the explanation more accessible to their young minds. Contingent reinforcement may be needed to ensure treatment adherence. For very young children, immediate positive reinforcement and incorporation of games may be indicated (Hirshfeld-Becker et al., 2010).

Graded in vivo exposure is the basis of CBT treatment for child anxiety (Hirshfeld-Becker et al., 2010). The cognitive aspect of CBT attempts to modify children's tendency to interpret ambiguous events negatively and to blame themselves for negative outcomes (Wilmshurst, 2015). One technique, cognitive restructuring, involves identifying the maladaptive thoughts that appear while imagining an anxiety-provoking situation. Once adept at identifying these thoughts, children can then learn to challenge them. Problem-solving sessions are useful when an anxiety-provoking situation has a realistic solution. Begin by brainstorming a list with the child and then selecting the most practical solution. This shifts the child's mindset from worrying to constructive thinking (Grover et al., 2006). After treatment is terminated, occasional booster sessions can be used as a way of reinforcing the skills and coping mechanisms learned during treatment (Gearing, Schwalbe, Lee, & Hoagwood, 2013).

Parents should be involved in the child's treatment whenever possible. They can be taught to model and reinforce coping strategies and to refrain from either criticizing or reinforcing anxious behaviors (Hirshfeld-Becker et al., 2010). Parents should be aware that their natural overprotectiveness of an anxious child may lead them to rescue their child from difficult situations, effectively preventing the development of inner resources (Steiner, 2011; Grover et al.,2006). Not only should parents be taught these techniques, but treatment should gradually shift responsibilities from the therapist to the parents (Manassis et al., 2014). When parents are themselves anxiety prone, they may need assistance to deal with their own anxiety before their child can benefit from CBT (Wilmshurst, 2015). However, therapists should avoid diluting the child's treatment with an excessive focus on parental anxiety (Feinstein, Fielding, Udvari-Solner, & Joshi, 2009).

Separation Anxiety Disorder

Children with separation anxiety disorder (SAD) experience intense distress around separation from caregivers. They might be consumed by worry about possible harm to the caregiver or a future separation, have difficulty going to school or sleeping alone, and may complain of physical symptoms when separation is imminent (e.g., headaches; APA, 2013). SAD is generally diagnosed between the ages of 6 and 18 and is commonly comorbid with GAD, depression, and somatic complaints. It is a possible precursor to various adulthood disorders such as depression, anxiety, panic attacks, and agoraphobia. Symptoms of SAD itself may resurface in adulthood under stressful conditions (Wilmshurst, 2015).

While a generalized anxiety treatment program can be used with SAD, disorder-specific treatment may be more effective (Mohr & Schneider, 2013). The essential aspects of treatment are graduated in vivo exposure and psychoeducation. Parental involvement is recommended as well, but can be omitted when impractical (e.g., with in-school treatment; Schneider et al., 2013). Therapists should tailor their techniques to the developmental stage of the child. For example, deep breathing can be explained to young children through the metaphor of blowing

into a balloon and allowing the air to escape. For a young child who has difficulty separating from caregivers before a session, separation can be gradual. For example, a parent might wait right outside the room during the first few minutes of a session, and once the child is comfortable with this, they may move to a waiting area down the hall (Grover et al., 2006).

Specific Phobia

Children with specific phobia have an unreasonable fear of being harmed by exposure to an object or place (e.g., specific animals or situations, natural phenomena, personal injury). This leads to avoidance of and escape from exposure situations as well as intense physiological responses when exposed, such as dizziness, breathlessness, and even fainting. In young children, the fear may be expressed through tantrums or clinging (APA, 2013; Wilmshurst, 2015). Specific phobias are often comorbid with anxiety disorders, mood disorders, and other phobias. They generally develop in childhood or adolescence (Wilmshurst, 2015).

The behavioral aspect of treatment involves graduated in vivo exposure, in conjunction with relaxation techniques, reinforced practice, and participant modeling (Wilmshurst, 2015; Steiner, 2011). Reinforced practice simply means rewarding participation in exposure to feared situations. In participant modeling, a model demonstrates staying calm during exposure before the child is exposed. Although live models may be most effective, video models have also been used successfully (Wilmshurst, 2015; Steiner, 2011), as well as puppets with younger children (Hirshfeld-Becker et al., 2010). The cognitive aspect of treatment targets children's exaggerated fearful beliefs. Children are encouraged to re-evaluate their beliefs after each exposure experience where the feared result does not occur (Ollendick et al., 2009).

Obsessive-Compulsive Disorder

Individuals with obsessive-compulsive disorder (OCD) experience unwanted obsessive thoughts as well as compulsive behaviors driven by these thoughts (Wilmshurst, 2015). To avoid the thoughts, the individual attempts to either ignore or suppress them or to alleviate them through some other thought or action (e.g., hand washing; APA, 2013). Approximately 1% of the population suffers from OCD, with slightly more females than males affected by the disorder. However, boys often experience the onset of OCD earlier in childhood than girls (Wilmshurst, 2015).

OCD can be treated with exposure and response prevention treatment, which is effective in both group and individual settings (Barlow, 2014). In this method of treatment, the client is exposed to a trigger and is prevented from engaging in either compulsive behavior or obsessive thinking. This contradicts their negative thinking using real-life experiences as evidence and decreases their anxiety through habituation (Lack, Storch, & Murphy, 2006). Clients whose fears include more abstract consequences (such as burning in hell) cannot be exposed to the feared object in session. In these cases, the client can simply imagine their fear (i.e., imaginal exposure). Children with OCD are prone to obsessing about performing perfectly on exposure exercises, which can make it burdensome and aversive. Therefore, therapists should advise them that repeated exposure is what counts, not perfect performance (Barlow, 2014).

The anxiety involved in exposure treatment often makes it difficult to gain the cooperation of child clients. Rewards can be used to increase some children's motivation. Some children may accept being told that the temporary discomfort is made worthwhile by the long-term gains. Parents can help motivate their children by avoiding "rescuing" them from their anxiety. Instead, parents should express their confidence in their child's ability to overcome the OCD despite the anxiety (Freeman, Garcia, and Coyne, 2008; Morgan et al., 2013, p. 189).

Depressive Disorders

Major Depressive Disorder

Children with major depressive disorder (MDD) consistently experience depressed or irritable mood or loss of interest or pleasure (APA, 2013). In addition, they may experience a variety of symptoms such as sleep problems, fatigue, difficulty concentrating, or feelings of excessive guilt. Suicidal ideation may also be present. These symptoms are severe enough to interfere with the child's important areas of functioning. Among children and adolescents, approximately 5% will suffer from MDD, although adolescent girls are diagnosed most frequently (Wilmshurst, 2015; Arnberg & Öst, 2014). Based on the child's developmental stage, MDD may manifest itself in distinct ways. Toddlers may show a lack of appetite or excessive head-banging; children may show poor academic and social functioning along with low self-esteem, vacillating between acting out and withdrawal; and adolescents tend to show more melancholic and suicidal tendencies. Anxiety disorders are commonly comorbid with major depressive episodes, and sufferers run a high risk of developing additional disorders, e.g., bipolar disorder (Wilmshurst, 2015).

Various forms of CBT with depression have proven effective (Arnberg & Öst, 2014). CBT treatment for MDD focuses directly on combatting the cognitive distortions that maintain the depression including a negative bias. Negative thoughts are identified and tested for accuracy. This helps the child develop more flexible ways of thinking, allowing for more positive thoughts (Steiner, 2011). The behavioral aspect of treatment is especially important, as it combats the low motivation that is characteristic of depression. By beginning treatment with a practical behavioral task, clients can begin to feel a forward momentum that can increase motivation. Social avoidance is a good target for this work, as the rewards of an enhanced social life can be highly motivating (Ahmed & Lawn, 2012). Psychoeducation can address parents' self-blame, as well as their difficulty disciplining a vulnerable child with depression. Realistic expectations should also be established regarding the course of MDD and the effects of treatment (Steiner, 2011).

When suicidal behavior is involved, the child's safety is paramount. Sessions should therefore be conducted in a context that allows for crisis intervention (including possible hospitalization). Further, the therapist should be trained and available to deal with suicidal crises whenever they may surface—even beyond session times. Suicidal clients benefit from therapy which restructures negative beliefs, encourages assertive communication, and facilitates creative problem solving. If the family exhibits dysfunctional patterns, addressing them can contribute to treatment success as well (Shaffer & Pfeffer, 2001).

Bipolar and Related Disorders

Bipolar I Disorder

Individuals with bipolar I disorder (BD) experience episodes of extreme moods, including depressive, manic, and hypomanic episodes, which interfere with normal functioning. Depressive episodes are identical to those of major depressive disorder; manic episodes are characterized by elevated or irritable mood and heightened energy and activity. Hypomanic episodes are shorter in duration and are not severe enough to impair normal functioning (APA, 2013). Child and adolescent onset bipolar disorder tends be more severe than later-onset BD and tends to rapid cycle between moods. Children with BD may have an illogically inflated self-esteem and experience problems with sleep due to their inability to relax their high level of functioning. High motor activity (e.g., multitasking) and risky sexual or financial behavior (e.g., theft) may be present. Care should be taken not to confuse BD with ADHD, as many symptoms overlap; the symptoms of elation and grandiosity appear to be the distinguishing factors between the two disorders (Wilmshurst, 2015).

In conjunction with medication to stabilize mood, cognitive behavioral techniques are used to improve emotional functioning (Steiner, 2011). Although controlled studies around CBT with bipolar disorder are scarce, Pfennig et al. (2014) reported success using a treatment that incorporated stress management, problem-solving strategies, and elements of mindfulness-based therapy.

Mindfulness involves nonjudgmental observation about one's own thoughts and experiences (Deckersbach, 2014). West et al. (2014) successfully used a group therapy model involving parents and children in separate groups. Sessions focused on cognitive restructuring, improving self-awareness, social and communication skills, and developing coping and problem-solving skills.

Psychoeducation should be provided with the goal of reducing the family tension that often accompanies BD treatment (Steiner, 2011). Families need to be taught coping skills such as identifying and responding when an episode is imminent. Parents need support in ensuring that the patient is medication compliant; a difficult task considering the medication's uncomfortable side effects (Barlow, 2014; Wilmshurst, 2015). Patients and families are also taught communication and problem-solving skills, such as repairing family relationships after an episode (Barlow, 2014; Steiner, 2011).

Trauma- and Stress-Related Disorders

Post-traumatic Stress Disorder

Children who present with post-traumatic stress disorder (PTSD) have been exposed to death, serious injury, or sexual violence, either in person by living this vicariously through a loved one. These youth may have flashbacks or nightmares about the event and may experience distress when exposed to cues that remind them of the trauma. Their overall mood and thinking may become negative or detached, and they may exhibit hypervigilance, e.g., overreaction to minor disturbances (APA, 2013).

It is important to note that children of different ages express PTSD symptoms in unique ways. School-age children may be restless and have difficulty concentrating during class and may overreact to loud sounds such as school bells. Preschool children may show separation anxiety, irritability, tantrums, and sleeping problems. Adolescents may engage in more risky or aggressive behavior and demonstrate survivor's guilt more often than younger children. Knowledge of the warning signs are essential, as PTSD is often missed by caregivers and educators who are inclined to minimize a trauma's effect on the child. Abused children may contribute to this situation by hiding the trauma to protect their parents, thus enabling other adults to deny its seriousness. A recent prevalence report of PTSD among adolescents cited numbers as high as 3.7% of boys and 6.3% of girls suffer from this disorder (Wilmshurst, 2015).

A variety of techniques are commonly employed in trauma treatment. Building a verbal narrative about the trauma allows the child to move on by transforming the memory of the trauma from a sensory memory to a verbal memory. This makes cognitive control possible, which can inhibit the intrusive emotional and physiological symptoms (Steiner, 2011). Moreover, organizing the trauma narrative can allow the individual to rebuild a sense of overall cognitive organization (Peri & Gofman, 2014). Building this narrative is most helpful with anxious and depressive symptoms, whereas externalizing symptoms are best addressed using additional parenting training sessions (Deblinger, Mannarino, Cohen, Runyon, & Steer, 2011). Thus, it is important to identify the most pressing concerns before choosing a treatment course. Graduated in vivo exposure to reminders of the trauma helps the youth build a tolerance for anxiety symptoms. Cognitive restructuring is used to develop more positive and less self-blaming ideas about the trauma. Training in emotional regulation as well as social and communication skills can help children manage and express trauma-related emotions in adaptive ways (Steiner, 2011).

Treatment can be made more child-friendly using various modifications. The child can com-

municate the difficulty of a task using a fear thermometer. Self-calming skills such as diaphragmatic breathing and progressive muscle relaxation may help the child feel more confident in therapy as well as in dealing with trauma reminders in the child's daily life. With very young children, art and play are used instead of words to express the trauma. Finally, helping the child to identify enjoyable and relaxing activities will help them self-regulate even outside of the therapist's office. To gain cooperation from parents, they should be provided with a clear rationale for exposing children to traumatic stimuli during treatment. Parents should also be trained to discuss traumatic experiences with the child as well as deal with their symptoms in a supportive and therapeutic way (Steiner, 2011).

Feeding and Eating Disorders

Bulimia Nervosa

Adolescents with bulimia nervosa (BN) are caught in a repetitive cycle of overeating followed by attempts to prevent weight gain. The disorder can present as the purging type (e.g., self-induced vomiting or laxatives) or non-purging type (i.e., fasting or excessive exercise). Youth with BN tend to evaluate their own worth largely on the basis of their own shape and weight. They purge in a misguided attempt to control body weight and binge when they interpret a minor lapse of self-control as a complete lack of self-control (APA, 2013; Barlow, 2014). BN is ten times more likely to occur in females than in males. Patients tend to keep within the average range of weight, with some fluctuations. Some will eventually be diagnosed with anorexia nervosa, while others may dispense with attempts to prevent weight gain and eventually be diagnosed with binge-eating disorder (Wilmshurst, 2015).

It is essential that the adolescent immediately begin keeping a food log with a careful record about binging and purging episodes. However, adolescents may lack the necessary commitment at first. If this is the case, early sessions can be devoted to beginning the log together, eventually transferring the responsibility to the adolescent. When providing a treatment rationale, keep in mind that adolescents often are unconcerned about the long-term effects of binging and purging. To engage the adolescent in treatment and show respect for their point of view, it is helpful to discuss negative consequences that they consider relevant, such as having to purge during a party with their friends (Lock, 2005).

After establishing a therapeutic alliance and record-keeping habits, the next stage should target the thought processes underlying BN. This includes beliefs about self-worth and fears of specific foods. Additionally, the adolescent should be encouraged to have more structured and healthy eating habits. Parents can be enlisted to help their child deal with anxiety, as long as the child is not opposed to this (Lock, 2005). The final stage focuses on relapse prevention. Adolescents should be provided with a realistic perspective about the possibility of a repeat episode. This will allow them to plan ahead to prevent a one-time relapse from leading to a full resumption of symptoms (Lock, 2005).

Anorexia Nervosa

As in bulimia nervosa, patients with anorexia nervosa (AN) tend to overvalue body shape and weight and the ability to control them. However, in AN, the psychopathology leads to more strict dieting without the compensatory eating, leading to extreme weight loss (Barlow, 2014). Lock and Le Grange (2013) discuss a family-based approach that has shown efficacy with adolescent AN. Briefly, parents should not be blamed for the illness; instead, they should be informed that their commitment is vital to the process of "refeeding" their child. This includes prevention of dieting, purging, and other symptoms of AN. Therapy is provided separately to parents and the affected child. The ultimate goal is to transfer all responsibility to the parents (Lock & Le Grange, 2013; Steiner, 2011).

Binge-Eating Disorder

In binge-eating disorder, the individual consumes a larger amount of food than is normal in a short period of time and does so repeatedly (APA, 2013). Binge episodes are most commonly triggered by negative affect. The episodes are characterized by a sense of loss of control and result in feelings of shame, guilt, or self-disgust. The disorder typically begins in adolescence to young adulthood (Wilmshurst, 2015).

Binge-eating disorder often leads to obesity. This is the primary reason why individuals seek treatment for the disorder (Lemberg & Cohn, 1999). The target of treatment is to break the cycle of food deprivation and subsequent binge eating. The guidelines used include eating in only one place, controlling the rate of eating, and clearly delineating the foods to be eaten during each meal. A behavioral group treatment has the additional benefit of providing support from other group members. The cognitive aspect of treatment follows a similar direction as that of bulimia nervosa in addressing the distorted beliefs regarding body weight and self-worth (Barlow, 2014).

Externalizing Disorders

Attention Deficit Hyperactivity Disorder

Attention deficit hyperactivity disorder (ADHD) is a psychiatric disorder affecting between 3% and 7% of the general population in the United States (Stein et al., 2009). ADHD is widely regarded as a persistent and biologically based disorder characterized by significant impairment in the area of executive functioning that persists into adulthood (Barkley, 1997, 2006). The core features of ADHD are inattention, impulsivity, and hyperactivity that impair functioning. There are three subtypes of ADHD: the predominantly hyperactive-impulsive type, the predominantly inattentive type, and combined type (APA, 2013). Children who suffer from ADHD often demonstrate significant impairment in their ability to sustain attention to academic tasks, delay gratification to meet long-term goals, understand consequences, plan, organize, and inhibit excessive motor activity (Hinshaw, 2006). Consequently, the above deficits may manifest as lost or misplaced homework assignments, disorganized backpacks, messy lockers, and misplaced folders for managing materials. Planning for long-term assignments, projects, and tests is also impacted markedly (Booster, DuPaul, Eiraldi, & Power 2012; DuPaul & Stoner 2003; Evans et al., 2005b; Langberg, Epstein, Urbanowicz, Simon, & Graham, 2008). Other peripheral features common to ADHD are inadequate social skills and peer relationships, low frustration tolerance, and strained family relationships (Rapport, Chung, Shore, & Isaacs, 2001).

A primary reason ADHD can be challenging to treat is because it is often comorbid with other psychopathology; most commonly anxiety or mood disorders, disruptive behavior disorders, and substance use disorders. These youth are also at risk to experience significant difficulties in the areas of academic performance and interpersonal skills (American Academy of Child and Adolescent Psychiatry [AACAP], 2007; Barkley, 2006).

Variations of cognitive behavioral therapy (CBT) were introduced in the early to mid-1980s to address the myriad of self-regulatory and cognitive deficits associated with ADHD. These interventions focused on teaching children problem-solving and specific strategies such as self-monitoring, modeling, role playing, self-instruction, self-reinforcement, and generating alternatives in decision-making situations (e.g., Abikoff & Gittelman, 1985; Hinshaw, Henker, & Whalen, 1984). However, a comprehensive review of literature demonstrates both the efficacy of behavioral interventions and the limitations of cognitive behavioral interventions in directly treating central ADHD impairments (Abikoff, 1991; Kazdin, 1997; Pfiffner, Barkley, & DuPaul, 2006; Schultz, Storer, Watabe, Sadler, & Evans, 2011). The limited impact of CBT reveals that the problems with attention, motoric activity, and impulsivity stem primarily from weakness in pre-verbal, neurologically based processes (Hinshaw, 2006). Therefore, these symptoms are inappropriate targets for CBT

interventions, which focus on the identification of cognitive distortions and interpretation of information processing (Kendall & Braswell, 1993). In fact, CBT outcome studies concluded that "there is little empirical support for its clinical utility with children with hyperactivity" (Abikoff, 1991, p. 205), a conclusion which was reaffirmed in a recent meta-analysis of CBT outcome studies for children with ADHD (Washington State Institute for Public Policy, 2012).

Children with ADHD need support in multiple environments including school, home, and the community. By the time a youth enters treatment, there has most likely been repeated efforts to support the child with their inattentive, hyperactive, and impulsive behaviors. Parents and teachers may feel frustrated, which places them at risk for developing cognitive misattributions about the child, such as believing the child's behaviors are purposeful or that even with interventions the behaviors will continue. The child is also at risk for developing cognitive distortions about himself and his relationships with peers and adults (Ostrander & Herman, 2006).

If misattributions surface, CBT can be used with the adults who are directly assisting youth with ADHD to stay focused and committed to the behavioral interventions they are applying (Kendall & Braswell, 1993). Parents and teachers can be taught to abandon their negative beliefs and replace them with more positive and accepting thoughts which will ultimately be favorable for the youth with ADHD and themselves (Anastopoulos, Rhoads, & Farley, 2006). Cognitive interventions can also be beneficial for children and adolescents who engage in negative self-talk, such as, "I am lazy" or "I am stupid." These youth can be taught about the impact of the disorder on academic and social performance producing interest in changing maladaptive patterns (Hinshaw, 2006; Ramsay & Rostain, 2008).

In a recent case study (Levine & Anshel, 2011), CBT strategies were implemented in a treatment design that included the parent, teacher, and child-focused interventions for an 8-year-old male with ADHD. The treatment goals for this youngster included providing cognitive and behavioral interventions to improve the child's behavior and academic participation and to address his inattentive and noncompliant behaviors at home. Both the teacher and the parents were trained in behaviorally based interventions for ADHD. Lastly, the child would participate in CBT-based counseling sessions to provide psychoeducation about his disorder, to address sad affect and anxiety about peer rejection, and to augment his motivation to participate in his treatment. The outcome proved positive as there was an increase in targeted behaviors, such as an increase in homework completion and a decrease in peer conflicts.

Although CBT has its limitations for the treatment of core symptoms of ADHD (Abikoff, 1991), parent and teacher cognitions play an integral role in the motivation and commitment to treatment. Further, cognitive interventions can be used to engage the youth in discussion about their self-image, self-esteem, and comorbid conditions such as anxiety and depression (March et al., 2004a, 2004b). Therefore, CBT can be a useful intervention for auxiliary treatment partners as well as the youth themselves.

Other Behavioral Disorders

Oppositional defiant disorder (ODD) is one of the most commonly encountered clinical disorders in children and adolescents. It is marked by a recurrent pattern of developmentally inappropriate levels of negativity, defiant, disobedient, and frequent hostile behavior toward authority. Behaviors commonly associated with ODD include temper tantrums, persistent stubbornness, resistance to directions, unwillingness to compromise, deliberate and or persistent testing of the limits, and verbal aggression. These behaviors are almost always seen in the home, with those the child knows well. It is likely the child also suffers from low self-esteem, mood lability, low frustration tolerance, and swearing (APA, 2013). ODD is frequently comorbid with other psychiatric conditions and often precedes the development of conduct disorder (CD), substance abuse, and other severe delinquent behaviors (AACAP, 2007).

Treatment of ODD can be considered multifaceted when there are comorbidities such as

ADHD and mood disorders. Data suggest that approximately 65% of children diagnosed with ADHD have comorbid ODD and that over 80% of children diagnosed with ODD have comorbid ADHD (Green et al., 2002). Other disorders such as major depression are seen in 70% of children with ODD, while bipolar disorder has a staggering 85% comorbidity rate with ODD. It is believed that children who meet criteria for a mood disorder are at an elevated risk for the development of conduct disorder (CD) (Green et al., 2002).

Adolescents with CD are often difficult to conceptualize and treat effectively. Many of these youth come from deprived environments with multiple stressors and a history of physical, emotional, and sexual abuse and/or neglect. As a group, CD disordered youth present with complex and recurrent behavioral problems, most notably aggression, bullying, violence, intimidation delinquency, rule violations, recklessness, property destruction, disregard for others, substance abuse, sexual abuse, and other disruptive and antisocial behaviors (Kazdin & Weisz, 2003). The prevalence rate for CD is between 6% and 16% for males under the age of 18 and is one of the most frequent disorders diagnosed in outpatient and mental health programs. Furthermore, 80% of these youth will most likely meet criteria for psychiatric disorders in the future (Kazdin & Weisz, 2003).

Treatments for children and adolescents with externalizing disorders such as ODD, ADHD, and CD often resort to the use of physical restraint, seclusion, and medication-based restraint. However, there is growing evidence that restraint and seclusion will often exacerbate aggressive behaviors (Denckla, 1996) and should not be used. Evidence-based treatment typically involves the demands of multimodal treatment, involving psychosocial and sometimes medication therapy (AACAP, 2007).

Both clinical and school systems are increasingly motivated to use evidence-based interventions to address youth's social, emotional, and behavioral needs. Although many empirically supported programs vary according to format, e.g., individual vs. group, different developmental stages, and the degree of parent/child involvement; there are several reoccurring themes, such as operational conditioning and contingency management. The use of rewards for positive behavior and consequences for negative behavior is often used. Additionally some programs reinforce children's engagement in positive goal setting (Lochman, Wells, & Lenhart, 2008), which promotes awareness of behavior. Interventions also typically include ways for the child to increase their emotional awareness and strategies to reduce and manage their anger. Some programs will include relaxation training as a way for aggressive children to reduce negative feelings in the moment and for future events (Feindler & Ecton, 1994).

Aggressive children often have deficits in their problem-solving and social skills Problem solving is viewed as a skill that can be taught and practiced and is a way for children to learn about the consequences for the choices they make (Kazdin, 2010). Kazdin's (2010) problem-solving skills training emphasizes teaching, practice, and reinforcement of pro-social problem solving. In addition, social skills, such as making and keeping friends, negotiating, and conflict resolution, are all important goals for intervention, as children with conduct issues often struggle in these areas. Further, adequate pro-social skills are associated with academic achievement (Caprara, Barbaranelli, Pastorelli, Bandura, & Zimbardo, 2000). Therefore, addressing the social, emotional and behavioral needs of the youth may improve academic functioning.

In comparison to single-component programs, multicomponent interventions lead to greater improvement and a reduction of aggression (Lochman, Salekin, & Haaga, 2003). Kazdin's (2010) *Parent Management Training*, Webster-Stratton and Reid's (2010) *Incredible Years*, and Zisser and Eyberg's (2010) *Parent Child Interaction Therapy* are all excellent programs that train parents to reinforce pro-social and compliant behavior. These programs stress the use of effective commands, ignorance of minor disruptive behaviors, and delivery of effective consequences.

In addition to behavior management training, these programs often address how to deal with stress, finding social support, parent involvement in schools, and how to effectively communicate.

Models known as parent training (PT) and behavioral family therapy have a primary focus on adjusting the way parents interact and ultimately discipline their children, as this is seen to impact the development of oppositional behavior and problematic parental interactions. Skills that are typically taught to parents are positive attending, use of appropriate commands, contingent attention and reinforcement, and the use of time-out procedures (McMahon, Wells, & Kotler, 1998). However, alternative models have contributed the thought of placing greater emphasis on the cognitive factors underlying ODD rather than on the behavior (Crick & Dodge, 1996; Kendall, 1985, 1991). These models focus on the cognitive deficiencies and cognitive distortions of oppositional and conduct disordered youth. In the above two models, neither truly addresses the reciprocal adult/child process which precipitates the aggressive behavior in a child (Greene, Ablon, & Goring, 2002).

A cognitive behavioral model of intervention known as *collaborative problem solving* (CPS; Greene et al., 2002) differs from many PT programs in its emphasis on facilitating the parent/child problem solving instead of teaching the child how to comply with adult requests and directives. It also contrasts from other cognitive behavioral models of intervention in its emphasis on assisting adults and children to develop skills to resolve issues of concern collaboratively. CPS is a manualized treatment program; however, session content and duration are not circumscribed (Greene et al., 2004).

One commonly held belief is that behaviorally challenging children often choose not to comply with adult expectations, because they have learned that disruptive behavior results in personal gains. These gains could take form such as getting one's way, receiving attention, or avoiding work. The CPS model, in contrast, conceptualizes childhood externalizing behaviors as the product of delayed cognitive skills in the broad domains of problem solving, flexibility, and frustration tolerance. In the CPS model, adults will identify a child's cognitive-skill deficits and then assist the child to build these skills through a process of collaboratively solving problems to find solutions that are mutually satisfactory (Pollastri, Epstein, Heath, & Ablon, 2013).

The aim of the CPS approach is to help the adults understand the cognitive factors that may be contributing to angry outbursts, and tantrums, low frustration tolerance, and poor adaptability. Subsequently, they become aware of the strategies for handling these unmet expectations. Parents are taught to recognize the impact of the adult/child interactions and become proficient at resolving conflict and defusing potentially conflictual situations where emotional outbursts are likely (Pollastri et al., 2013).

CBT has been utilized widely in treatment programs for youth who are behaviorally disordered, but there are limits to the effectiveness of CBT in youth that are psychologically complex (Young, Klosko, & Weishaar, 2003). Therefore, an advanced form of cognitive behavioral treatment called "Mode Deactivation Therapy" (Apsche & Ward-Bailey, 2004) was developed to address multiple issues of conduct-disordered youth. Mode deactivation therapy (MDT) has been utilized in treating adolescents who engage in sexually harmful behaviors as well as adolescents with severe mental illness. MDT is an evidence-based treatment that merges key elements from Beck's theories of "modes" (Beck, 1996), traditional CBT and schema therapy (Alford & Beck, 1997; Beck & Freeman, 1990), dialectical behavior therapy (Linehan, 1993), and functional analytic behavior therapy (Kohlberg & Tsai, 1993; Nezu, Nezu, Freeman, & Haynes, 1998).

MDT has been shown to be an effective treatment to an array of adolescent disorders (Apsche, Bass, & Siv, 2006) including emotional dysregulation (Apsche & Ward-Bailey, 2004), behavioral dysregulation (Apsche, Bass, & Murphy, 2004), physical aggression (Apsche, Bass, & Houston, 2007), sexual aggression (Apsche, Bass, Jennings, Murphy, Hunter, & Siv, 2005), and many harmful symptoms of anxiety and traumatic stress (Apsche & Bass, 2006). MDT family

therapy has also shown promising effects in the reduction of family discord in case studies (Apsche & Ward, 2004). Subsequently, MDT has been shown to be efficacious as compared to treatment as usual in treating families with a myriad of complex problems (Apsche & Bass, 2006) and in maintaining treatment effects through 2 years of progress monitoring (Apsche, Bass, & Houston, 2007).

MDT shares the basic principles of CBT (Young et al., 2003). MDT concurs that atypical behavior originates from dysfunctional schema that trigger "modes"; yet, the approach is drastically different in how it corrects this schema. Unlike CBT, MDT does not directly challenge irrational thoughts or disrupt cognitive distortions; instead these core beliefs or schemas are consistently validated as legitimate (Apsche & Bass, 2006). The schema are then "balanced" through a collaborative effort between the therapist and the youth; this is a process to deactivate the maladaptive mode responses (Apsche & Bass, 2006).

MDT also includes a series of mindfulness exercises that are specifically intended for adolescents. There is a client workbook that is designed to allow the youth to practice the skills they learn to reduce anxiety, build trust and increase their commitment to therapy. As the youth continues to practice these exercises, they are able to increase their awareness of their own emotions and feelings. The MDT mindfulness is a critical element to this treatment (Apsche & Bass, 2006).

In a study concerning the treatment of adolescent males with conduct disorder, MDT, CBT, and social skills training (SST) were compared. The results indicate that MDT achieved superior results to traditional CBT and social skills training (SST) in reducing both physical and sexual aggressions in conduct-disordered youth in a long-term residential treatment environment. Although CBT and SST showed effectiveness in the reduction of physical aggression, only MDT showed marked reduction rates of sexual aggression. This suggests that the modifications of the cognitive behavioral treatment used in MDT may be especially useful in the treatment of unique and complex clinical presentations of these behaviorally disordered youth (Apsche & Bass, 2006).

In conclusion, based on data from a recent meta-analysis in which a variety of problems were examined, CBT demonstrated the strongest efficacy for anxiety, followed by depression and bipolar depression. CBT was also found to be considerably more effective than other treatments for bulimia (Hofman, Asnaani, Vonk, Sawyer, & Fang, 2012). In contrast, there is limited evidence that supports CBT is efficacious in the treatment of ADHD (Antshel, Faraone, & Gordon, 2012), and many of the other externalizing behavioral problems respond best with multicomponent programs and interventions which leads to a reduction of behavioral problems and aggression, as well as maintenance of gains (Lochman et al., 2003).

Conclusion and Future Directions

In this chapter, we have outlined several techniques commonly used in CBT, as well as treatments that have been adapted from the CBT model. A goal is to increase the effectiveness of any treatment; therefore, we will spend our concluding remarks on ways to enhance the effectiveness of CBT and CBT-based treatments.

There are cases where CBT has been ineffective in combatting the presenting problems in treatment. If this occurs, an adjunctive pharmacological treatment may be helpful and beneficial. For example, newer antidepressant agents have been shown to enhance the treatment's effectiveness when used in conjunction with CBT (Hides, Samet, & Lubman, 2010). The addition of booster sessions aimed to monitor and maintain progress may also improve the long-term outcomes for CBT (Hides et al., 2010), and mindfulness-based cognitive therapy (Carroll & Rounsaville, 2007) has been shown to be efficacious in the treatment of anxiety and mood disorders (Hofmann, Sawyer, Witt, & Oh, 2010).

Lastly, in the age of mass Internet use, we would be remiss without discussing the use of internet based CBT. A recent meta-analysis reviewed literature on computer-based treatments

for depression and concluded a superior effect size for therapist-supported computer-based interventions and a moderate effect size for programs that did not have a therapist supporting the user (Richards & Richardson, 2012). Given the prevalence of various disorders and the lack of suitably trained CBT practitioners, it is surprising that computerized treatments are not used more widely (Richards & Richardson, 2012). Furthermore, today's children and adolescents are highly computer literate, making computers a natural treatment medium at a lower cost (Abeles et al., 2009). As these lower-cost treatments mature in credibility, they hold the promise of meeting the needs of many more children in the near future.

Case Study

Emily is an 11-year-old girl who was referred to a psychologist for her intense emotional, behavioral, and physiological reactions to upcoming state school. Her parents reported that on those days where her teachers were to work on preparation for these exams (in both math and English/language arts), Emily stayed in her bed, refused to leave the house, and according to her parents "wailed hysterically" to the point where the family's landlords had to check in to ensure that everyone was all right.

Her parents reported that Emily was a bright young girl but she had a tendency to keep to herself. They did not describe her as being shy, but she did have a tendency to "follow her own rules" and never developed a consistent group of friends in school or in any of her extracurricular activities. Her teachers had always described her as a slightly below average student academically (her parents corroborated that it was a "nightmare" to get her to do homework), but quite bright when engaged in a one-on-one situation. They used phrases such as "spacey" and "in her own world" to describe Emily. She also tended to demonstrate tendencies to blurt out answers in class.

After an extensive period of data collection, from Emily's parents, her current and previous year's teacher (who had worked as her tutor this year), and Emily herself, the psychologist felt that two diagnoses were warranted: *attention deficit hyperactivity disorder*, *predominantly inattentive presentation*, and *social anxiety disorder* as per the DSM-5 guidelines (APA, 2013). As was mentioned before in this chapter, it was important to collect as much data as possible in order to view the problem holistically. While the presenting problem was the presence of symptoms that suggested a *social anxiety disorder*, the psychologist in this case felt that this anxiety was secondary to her primary diagnosis of *ADHD*, *predominantly inattentive presentation*.

There is evidence to suggest that the presentation of ADHD varies significantly among boys and girls (Gaub & Carlson, 1997) where girls will present with lower levels of hyperactivity and lower rates of externalizing difficulties. As such, the gender discrepancy between males and females diagnosed with ADHD may be due to a referral bias for more externalizing and hyperactive behaviors. Furthermore, there are higher rates of anxiety and mood disorders as well as learning disabilities found in females diagnosed with ADHD (Quinn, 2005). In this instance, Emily's learning challenges and her difficulties with the inhibition of her behaviors may have come to a head due to the excessive environmental demands of test preparation, which led to a complete shutdown in the form of excessive crying and refusal to go to school.

Emily's treatment focused on not just helping her directly, but working with the adults around her in order to structure her environment to make it conducive for her mental and behavioral health. First and foremost, Emily's parents were educated regarding the nature and course of Emily's diagnoses. They worked with the psychologist to structure their home environment in a way that was simultaneously supportive yet structured. Her parents were assigned readings from various websites and were encouraged to attend local support groups to reach out and find other parents whose children presented similar issues. In addition, Emily's parents were encouraged to speak to her school-based support team in order to

examine what supports might be available to her to help her academically as well provide accommodations for her state exams that might lessen her anxiety (e.g., alternative setting and/or extended time to complete her exam).

Emily was also educated as to the nature of her diagnoses, and what those diagnoses mean for her currently and in the future. In many respects, Emily was quite relieved to discover that her issues had some sort of real cause (and not because she was a "space cadet" as someone in her school had said a few years ago). This understanding prompted her to explore the issue further with handouts and websites that the psychologist recommended to her.

Once school supports were put into place, and a structured home environment was instituted in order to help Emily attempt and complete her homework and long-term projects, the psychologist began to teach Emily relaxation strategies in the form of deep breathing and progressive muscle relaxation techniques (PMR; Masters, Burish, Hollon, & Rimm, 1987). In addition to teaching Emily these techniques, the psychologist also taught her parents these techniques as well, so that they could model them for her during times of crisis.

The psychologist also encouraged Emily to try to identify optimal times to use various relaxation strategies. Some strategies were not appropriate for whole classroom activities (e.g., one PMR technique encourages opening the mouth and sticking out the tongue as far as possible). The optimal times to practice these techniques and decide which ones were more effective were during one of the many mock practice exams. Ultimately, Emily selected a combination of deep breathing just before the exam coupled with the systematic tensing and relaxation of her arms, hands, and legs during the exam as a relaxation cocktail that worked for her. This seemed to reduce her anxiety significantly during the actual state exam.

Emily has been successful so far in therapy (this is an ongoing case). A critical key to this success was helping Emily and her parents understand that the diagnoses that she carried were not issues to be cured of; rather, they were representative of chronic issues that would flare up during various situations. The psychologist and the family identified future periods in Emily's academic and personal life where such issues might occur and laid out plans to deal with these potential problems, such as future exams and transitions to high school. Now that some of her immediate concerns have been addressed, Emily will need help to develop appropriate peer relationships that can serve as a resource when she experiences future stressors.

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Behavioral Interventions

32

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Interventions for the treatment of attention-deficit/hyperactivity disorder (ADHD), oppositional defiant disorder (ODD), and conduct disorder (CD), collectively referred to as disruptive behavior disorders (DBDs), are a high public health concern. These disorders share common core features (i.e., poor behavioral inhibition and emotion regulation) and invariably manifest behaviorally as poor compliance with adult instructions, rules, or expectations (Olson, Sameroff, Kerr, Lopez, & Wellman, 2005; Frick, Barry & Kamphaus, 2010; Blair, White, Meffert & Hwang, 2014). These behavior problems are distressing for adults and represent a significant proportion of early childhood mental health problems (Brestan, Eyberg, Algina, Johnson, & Boggs, 2003; Scahill & Schwab-Stone, 2000; Nock, Kazdin, Hiripi & Kessler, 2007). Moreover, children diagnosed with one or more behavior disorders face increased risk for poor mental health, substance abuse, delinquency and legal challenges, and impaired social relationships in adolescence and adulthood (Hartman, Stage & Webster-Stratton, 2003; Rowe, Costello, Angold, Copeland, & Maughan, 2010; Sibley et al., 2011; Mordre, Groholt, Kjelsberg,

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Sandstad, & Myhre, 2011; Burke, Rowe, & Boylan, 2014). Given the prevalence, costs, and outcomes associated with DBDs, effective treatment is critical to altering poor outcomes.

Herein we focus our attention on both well-established and emerging interventions for the treatment of DBDs. We adopt a classification system employed by Evans and colleges (2014) that categorizes interventions into behavioral management intervention (interventions that teach individuals other than the patient skills to modify the patient's behavior in context) or training interventions (interventions that teach/focus more directly on the target patient to improve their skills). Behavior management interventions primarily include behavioral parent training, behavioral classroom management, and behavioral peer interventions—all of which are primarily used with preschool through school-age youth with ADHD, with emerging evidence that these interventions can be effective in addressing the functional impairments of adolescents with DBDs. Training interventions include social/cognitive problem-solving interventions and organizational skills training. Additionally, we briefly discuss emerging interventions for the treatment of DBDs, specifically the Collaborative and Proactive Solutions intervention, multiple family groups, and neurocognitive interventions. We close with future directions in the treatment of DBDs.

Behavior Management

Behavioral Parent Training

Behavioral parent training (BPT) is an evidence-based psychosocial intervention strategy for preadolescent children who engage in developmentally inappropriate levels of disruptive behaviors or who meet criteria for DBDs (Eyberg, Nelson, & Boggs, 2008; Evans et al., 2014). As the name suggests, BPT interventions are designed to teach parents and other caregivers how to functionally apply strategies (i.e., positive reinforcement and punishment) based on principles from behavioral psychology (operant conditioning and social learning theories) to change maladaptive child behaviors (Moreland, Schwebel, Beck, & Wells, 1982). In other words, parents learn to strategically encourage and teach appropriate child behaviors and discourage inappropriate behaviors. Parenting is a well-established mediating factor in the development of oppositional and conduct problems in youth (Patterson, 1982; Dishion & McMahon, 1998). Importantly, although parenting is not implicated in the development of ADHD per se, parenting is a notable factor in the development of comorbid ODD, CD (Chronis et al., 2007; Pardini & Fite, 2010), depression (Ostrander & Herman, 2006), and anxiety (Kepley & Ostrander, 2007; Pfiffner & McBurnett, 2006) in youth with ADHD. As such, targeting parenting behaviors is necessary when working with youth with DBDs.

The efficacy of BPT in improving parenting behavior, opposition, and conduct problems has led to universal inclusion of BPT in practice parameters for children with ADHD, ODD, and CD (e.g., Evans et al., 2014; Steiner & Remsing, 2007; American Academy of Child and Adolescent Psychiatry, 2007a, b; American Academy of Pediatrics, 2011; Steiner & Dunne, 1997). Moreover, there has been extension of the BPT model to adolescents with ADHD and comorbid DBDs, focusing on both behavior management implemented by the parent and the use of problem-solving communication training for both parents and adolescents (Robin, 2015). In fact, behavioral interventions are considered the first-line treatment option for addressing functional impairments in adolescents with ADHD (Sibley, Kuriyan, Evans, Waxmonsky, & Smith, 2014). The following review describes the development and core components of the BPT model.

Given their common origins and behavioral underpinnings, it is not surprising that the manualized BPT programs used to treat preadolescent disruptive behaviors overlap substantially in their procedural components. Most BPT models involve service delivery to an individual family, although group-based models have also been developed. Regardless of delivery mode, programs typically begin with teaching parents specific

techniques to increase their responsiveness to child signals and strengthen the parent-child relationship (e.g., "Child-Directed Interaction" in *Parent-Child Interaction Therapy*, McNeil & Hembree-Kigin, 2011; "Child's Game" in *Helping the Noncompliant Child*, McMahon & Forehand, 2003). Strategies include allowing the child to choose the game and how to play; providing contingent praise for appropriate behaviors; using imitation, description, and paraphrasing of the child to enhance the parent's positive attention; and avoiding negative statements and unnecessary questions or instructions. For older school-age youth, positive attending to children's preferred activities are analogous methods that are employed.

The second phase of treatment (analogous to Hanf's second stage, described above) teaches parents how to give developmentally appropriate instructions, issue a single warning (if needed), provide contingent praise for compliance, and administer a brief time-out for noncompliance followed by reinstruction. Several programs also integrate a focus on parent monitoring and implementation of token reinforcement systems (e.g., Patterson, Reid, Jones, & Conger, 1975). Data collection on parent skill acquisition takes place throughout the intervention in most programs. Consistent with behavioral learning theory, clinic sessions involve extensive therapist modeling of the procedures described above, as well as in vivo parent practice with feedback. For adolescents, in addition to behavioral management procedures employed by parents, parents and adolescents learn communication skills, identify existing ineffective communications behaviors, and work toward resolving issues while systematically using effective communication skills (Robin, 2015).

Kaminski, Valle, Filene, & Boyle, 2008 conducted a systematic review and meta-analysis to examine what BPT model components (including those just described) best predicted positive response to treatment. Inclusion of a component to teach parents how to increase positive interactions with their children and teaching approaches that involved parents practicing skills directly with their children in session were both associated with larger treatment effect sizes for child behavior improvement (Kaminski et al., 2008). Similarly, inclusion of parent training on time-out administration and consistent discipline was also associated with larger treatment effects. Simply teaching parents about child development was not associated with improved outcomes.

Now that BPT is well-established as an efficacious treatment for DBDs, outcome studies are currently focused on decreasing barriers, improving access, and enhancing engagement to BPT for youth and their families. Despite the efficacy of BPT, families frequently fail to engage sufficiently in traditional BPT programs, as evidenced by decreased attendance, lower rates of homework completion, and higher rates of dropout (Chacko et al., 2013; Nock & Ferriter, 2005; Armbruster & Kazdin, 1994). There are a number of factors theorized to influence these poorer outcomes: parental beliefs that the child should be the target of effective treatment instead of the parent, as well as significant barriers to treatment including difficulties arranging time off work to attend sessions, problems with transportation, lower levels of social support, and issues related to child care during clinic visits, among other factors (Koerting et al., 2013).

Adaptations to traditional BPT have been successful in improving engagement in difficult-to-engage families. As an example, traditional BPT has been adapted to include explicit discussions of practical barriers and maladaptive cognitions related to treatment, improving social support and motivation during treatment, addressing parental depression, and more directly addressing problems related to parent- and child-related difficulties that arise during treatment, resulting in greater engagement to BPT in high-risk families (Chacko et al., 2009; Chacko, Wymbs, Chimiklis, Wymbs, & Pelham, 2012; Chronis-Tuscano et al., 2014; Kazdin & Whitley, 2004; Nock & Kazdin, 2005). Other adaptations have included modifying the parameters of traditional BPT. As an example, Fabiano et al., (2009; 2012) focused on teaching the utilization principles of BPT in the context of soccer games, which was hypothesized to be more engaging for fathers of youth with ADHD relative to just traditional

classroom-based instruction. Results suggested increased engagement relative to a traditional BPT program (Fabiano et al., 2009; 2012). Others have improved engagement to BPT through reducing the frequency of BPT sessions (Meyer & Kelley, 2007) or by utilizing technology (e.g., mobile phone applications) to further support parents when implementing BPT (Jones et al., 2013; McGrath et al., 2011).

Interestingly, while these adaptations appear to have clear benefits on engagement, there is a question as to the incremental benefit of these enhanced BPT interventions on outcomes (Chacko et al., 2013; Chronis-Tuscano et al., 2014; Evans et al., 2014; Kazdin & Whitley, 2004). As an example, some data suggest that there are very few differences between traditional BPT and enhanced versions of BPT on the rate of clinical significant outcomes (Chronis-Tuscano et al., 2014; Rajwan, Chacko, Wymbs, & Wymbs, 2014). We believe that next steps in this line of research should more carefully consider tailoring adaptations to BPT for those families who require support based on poor response to BPT (see Chacko et al., 2015 for a discussion).

Behavioral Classroom Management

DBDs are associated with academic and behavioral impairments in the classroom setting (American Psychiatric Association [APA], 2013; Van Lier, Muthén, van der Sar, & Crijnen, 2004). As such, behaviorally based treatment techniques for DBDs targeted at decreasing impairments in the classroom setting are comparable. Research supporting the implementation of classroom-based behavioral interventions for DBDs dates back to the 1960s (Pelham, Wheeler, & Chronis, 1998; see studies below). By 1998, behavioral interventions for ADHD and behavioral and cognitive-behavioral interventions for disruptive behaviors were classified as well-established treatments (Brestan & Eyberg, 1998; DuPaul, Eckert, & Vilardo, 2012; Evans, Owens, & Bunford, 2013; Eyberg et al., 2008; Pelham & Fabiano, 2008; Pelham, Wheeler, & Chronis). Studies of youth with DBDs suggest that classroom behavior management can lead to significant improvements in adherence to class rules, increased attainment of behavioral goals (Fabiano et al., 2010), and significant reductions in frequency and severity of ODD/CD behaviors (Kapalka, 2010; Fabiano et al., 2004, 2010). A recent meta-analysis of school-based treatment for children with ADHD found that these interventions yield moderate to large effects for both behavioral and academic outcomes and concluded that school-based interventions should be a first-line treatment for students with ADHD (DuPaul et al., 2012).

The focus of behavioral classroom management intervention is to understand the function of problematic school/classroom behaviors and to modify antecedents (i.e., factors that occur before the target behavior) and consequences (factors that occur after the target behavior) associated with the occurrence of identified target behaviors. Teachers are often supported in using antecedent-focused methods (e.g., classroom rules, effective commands), consequence-focused methods (e.g., labeled praise, time-out from positive reinforcement), and methods that target both antecedent and consequences (e.g., school-home daily report cards, token economy systems) to improve targeted behaviors in the classroom setting. Over the years, multicomponent programs that incorporate antecedent-based and consequence-based techniques have been developed for classroom settings to reduce disruptive behavior. For example, the teacher training component of the Incredible Years program has been found to significantly reduce problem behavior in the classroom when it is implemented in conjunction with child and/or behavioral parent treatment (Eyberg et al., 2008; Webster-Stratton, Reid, & Hammond, 2004). This program is for teachers of younger children, and it consists of training modules that include classroom rules and structure, differential attention, praise, incentives, time-out, and skill building. The Good Behavior Game is an example of an evidence-based group contingency incentive program developed for the classroom setting (Barrish, Saunders, & Wolf, 1969). This game involves dividing the class into two or more teams with

the goal of having the fewest classroom rule violations during a given period of time. The team with the fewest violations without reaching a predetermined limit earns privileges. Short-term and long-term improvements in child disruptive behaviors have been found for the Good Behavior Game (Kellam, Brown, Poduska, Ialongo, Wang, Toyinbo et al., 2008).

Importantly, effective implementation of behavioral classroom management procedures requires collaboration between teachers, school staff, parents, and therapist(s). Collaboration can be challenging given that each member of the team may have a different perspective with regard to treatment planning. Communication between each member of the team may also be challenging given that their roles with the child are typically managed in separate settings. It is therefore important for parents, as the key organizing adult in the child's life, to learn advocacy skills that will help them communicate their child's needs when there are significant barriers to behavioral consultation (see Chacko et al., 2015 for a discussion). Written documentation of classroom accommodations and behavioral management techniques that are implemented in the classroom can be incorporated formally into a Section 504 plan/Individualized Education Program or informally through e-mail/letters to ensure accurate communication of goals and treatment plans.

A lack of teacher training on evidenced-based practices for children with disruptive behavior problems may serve as another barrier to implementing behavioral classroom management procedures. The National Council for Accreditation of Teacher Education reports standards for school psychologists related to behavioral assessment and intervention, yet teacher competency in this area is not emphasized (Kellam et al., 2011; National Council for Accreditation of Teacher Education, 2008). This highlights a broader issue that there is deficient dissemination of evidenced-based practices for behavioral classroom management. Mental health professionals trained in these practices can offer consultations and training, but may not be able to observe and provide feedback on treatment implementation in longer term.

Behavioral Peer Interventions

Positive social relationships are an integral part of healthy development and reduce the negative impacts associated with peer rejection such as emotional disorders, delinquency, substance abuse, and other negative outcomes (Hoza, 2007a, b). Unfortunately, children with DBDs often have significant social impairments due to social skills deficits and/or child disruptive behavior (Cadesky, Mota, & Schachar, 2000; Hoza, 2007a, b; Kazdin, 1997; Ronk, Hund, & Landau 2011). Social skills training was developed to reduce negative social behaviors and increase prosocial behaviors in order to improve the interactions children have with their peers (Ronk et al., 2011).

Traditional social skills training (SST) consists of weekly, clinic-based sessions in a group format with didactic and role-play components with targeted children. These interventions have minimal or no effects on social impairments at the group level and may not generalize to real-world settings (Pelham & Fabiano, 2008). Conversely, behavioral peer interventions that consist of social skills training imbedded into naturalistic settings and the involvement of caregivers are now considered well-established treatments for children with ADHD (Evans et al., 2013) and probably efficacious treatments for children with ODD and CD (Eyberg et al., 2008).

The majority of the evidence in support of behavioral peer interventions for ADHD is from the summer treatment program (STP), a multicomponent, 8-week intensive day camp for children with ADHD in which behavioral modification is implemented in a recreational and classroom settings (Evans et al., 2013; Pelham et al., 2008; 2000; Pelham & Fabiano, 2008). Interestingly, a recent study conducted in a STP classroom setting suggested that the way in which teachers implement behavioral management techniques can improve peer acceptance in youth with ADHD. Mikami et al., (2013) compared a contingency management intervention to the Making Socially Accepting Inclusive Classrooms (MOSAIC) intervention.

The traditional contingency management intervention involved teachers awarding children points for prosocial classroom behaviors (similar to classroom procedures implemented in the STP). Points earned were publicly reviewed at the end of the activity by teachers and children traded in points for incentives. MOSAIC included the aforementioned contingency procedures with the exception that children received private feedback on their behavior and classroom rules were added to promote social inclusion of peers. Additionally, teachers were trained to assign collaborative activities and to encourage positive peer interactions by pairing children with similar interests and positively reinforcing social acceptance. Teachers were also instructed to emphasize the strengths of children who were observed to have behavior problems with the hope of preventing peer rejection. Results indicated that children who received MOSAIC, especially boys, had greater improvements in peer preference and more friendships than those who received the contingency management intervention.

Although the effects of the STP on social outcomes have been demonstrated (Pelham et al., 2008; 2000) and MOSAIC appears promising (Mikami et al., 2013), the extent to which replication of STP procedures can be conducted in more routine settings is debatable. As such, alternative, practically feasible methods are necessary. One such program is Parental Friendship Coaching (PFC; Mikami, Lerner, Griggs, McGrath, & Calhoun, 2010). In PFC, parents of school-aged children with ADHD learn effective methods to be social coaches for their children. The treatment consists of didactic, role-play, and playgroup feedback components. Session content includes teaching parents how to coach social skills (play skills and making friends) and how to organize and prepare for playdates to prevent conflict. Initial results have found that PFC can lead to improved child peer relationships (Mikami et al., 2010).

There is also evidence to support behavioral peer interventions for children with DBDs more broadly. The Incredible Years Child Training program, Dinosaur School, focuses on directly teaching and supporting young children's use of social-emotional-problem-solving skills. In this intervention, children watch video vignettes of real-world situations and discuss and practice problem-solving and social skills such as effective communication and cooperation. Furthermore, parents and teachers use contingency management strategies to reinforce the skills learned in the program at home and school, respectively (Webster-Stratton et al., 2004; Webster-Stratton, Jamila, & Beauchaine, 2011). The Dinosaur School has been found to increase children's social and emotion regulation, problem-solving ability, and social competence in young children at risk for or diagnosed with DBDs (Webster-Stratton et al., 2004; 2011).

Collectively, key aspects of effective behavioral peer interventions include contingency management procedures to help directly support social skills that children learn within natural contexts (e.g., classroom, playgrounds, home). Additionally, it appears that involvement of caregivers (i.e., parents and/or teachers) in implementing methods (e.g., coaching) and directly reinforcing social behaviors in children is necessary. In classroom settings, peer-wide contingencies that promote peer acceptance and inclusion should be included in behavioral peer interventions (Mikami et al., 2013). Although behavioral peer interventions have garnered empirical support as a well-established intervention for ADHD (Evans et al., 2014), the studies cited in support of this type of intervention were conducted in highly structured analogue contexts (Pelham et al., 2000; 2008; Mikami et al., 2013) or have used wait-list control comparison conditions (Mikami et al., 2010). As such, there needs to be further study of these approaches under more routine contexts, while still utilizing rigorous methods, to better determine the effectiveness of these approaches in improving social outcomes for youth. Given the importance of peer relationships in longer-term functioning of youth, identifying meaningful and feasible intervention approaches that can address social difficulties is critical.

Training Approaches

Social/Cognitive Information-Processing Interventions

Anger and aggression are hallmarks of DBDs, notably OD and CD. There has been a long history of training interventions that target the social/cognitive information-processing models (Crick & Dodge, 1994) that are often found to be disrupted in youth who exhibit anger and aggression. Youth with anger and aggression often perceive events to be threatening and attribute events to be provocative. These youth are more likely to focus on hostile cues in the environment, remember fewer potential cues, and perceive ambiguous events as hostile. Based on these perceptions and attributions, children form cognitive plans to respond to the perceived threat or provocation. These plans are often limited in range, relying heavily on aggressive behaviors, which are viewed by the youth as having an overly positive effect on the situation. Compounding these maladaptive processes, youth often have poor perspective of emotions in others, difficulties regulating their own emotions, and limited perspective of the impact of their behavior.

Although there are many interventions that utilize the social/cognitive information-processing model as a basis for intervention, we focus on problem-solving skills training (PSST; Kazdin, Esveldt-Dawson, French, & Unis, 1987a, b) and Anger Coping/Coping Power training (Larson & Lochman, 2002; Lochman, Wells, & Lenhart, 2008; Wells, Lochman, & Lenhart, 2008) given that these interventions are the two most well-studied social/cognitive information-processing model-based training interventions for youth with DBDs.

Broadly speaking, PSST and Anger Coping/Coping Power focus on improving emotional awareness, perspective taking, decision-making processes, and coping strategies (e.g., relaxation strategies). PSST is a clinic-based intervention, while Anger Coping/Coping Power are (primarily) school-based interventions. Youth are taught specific skill sin a step-by-step basis in either individual sessions with the therapist (PSST) or group sessions with peers (Anger Coping/Coping Power). Skills are taught through didactics, role-playing, therapist modeling, and reinforced practice. Homework assignments each week include children practicing skills with others such as their peers. Importantly, parent involvement in supporting youth when implementing the skills they learn is often an essential aspect of these interventions. A scaffolding approach is taken, whereby clinicians gradually provide less support as children successfully implement problem-solving skills of increasing difficulty during sessions and in real-world situations. A token economy system is used to reward children's practice of skills and to provide mild punishment for inappropriate behaviors.

Multiple randomized controlled trials have been conducted on PSST and Anger Coping/Coping Power. PSST has been found to improve oppositional and aggressive behavior as well as prosocial behavior relative to a wait-list control condition and a relationship-focused intervention, with maintenance of treatment gains over a 12-month follow-up period (Kazdin et al., 1987a; Kazdin, Bass, Siegel, & Thomas, 1989). Interestingly, effects of PSST are maximized when parents concurrently participate in behavioral parent training, resulting in significantly greater improvements as well as increasing the rate of clinical response in treated youth (Kazdin et al., 1987b; Kazdin, Siegel, & Bass, 1992).

Similarly, in randomized controlled treatment designs, Anger Coping has been found to be more effective than attention-control treatment in improving disruptive behavior at home and school (Lochman, Burch, Curry, & Lampron, 1984), with data suggesting longer-term effects on improving classroom on-task behavior (Lochman & Lampron, 1988) as well as reducing the risk of adolescent substance use outcomes (Lochman, 1992). Coping Power has also been shown to be efficacious. In a large school-based randomized controlled trial, Lochman and Wells (2004) found that the combined parent and child Coping Power program resulted in lower rates of delinquent behavior and substance use than the child-only Coping Power program and a wait-list

control condition. In a clinic-based study of youth wit;h DBDs, Coping Power was found to be more effective than services-as-usual treatment on improving aggression (van de Wiel et al., 2007), with 4-year follow-up data indicating greater reduction in some aspects of substance use for youth who participated in the clinic-based Coping Power program (Zonnevylle-Bender, Matthys, & Lochman, 2007).

Collectively, multiple RCTs have been conducted on interventions focused on improving the social/cognitive information-processing deficits in youth with DBDs, including studies of PSST and Anger Coping/Coping Power. It is important to note that these interventions are geared toward older school-age and middle school-age youth, given the relative advanced cognitive requirements of the program. It is important to note that these interventions require support from parents and/or teachers for maximal effects. This is not surprising given that the larger family and school context often plays a significant role in the development and maintenance of these problems (Patterson, 1992).

Organizational Skills Training

Youth with ADHD often have considerable and persistent difficulties with organization, time management, and planning (Barkley & Fisher, 2011). Not surprisingly, these skills are critical for being able to successfully navigate school, particularly as children get older. Older children are often more responsible for organizing their own school materials, recording assignments, and planning for and completing homework (Power, Werba, Watkins, Angelucci, & Eiraldi, 2006). Disorganization, poor time management, and planning can significantly disrupt these important, daily school activities, resulting in significant impairments at school and increased conflict at home. Although difficulties in organization, time management, and planning skills have been a long-standing concern for parents and teachers, it is only within the past 10 years that specific efforts have been made to train students with ADHD to improve these skills.

Abikoff and colleagues (2012) developed an organizational skills training intervention for school-age youth with ADHD. The organization skills training intervention focused on teaching children to use new tools and routines to record assignments, organize school materials, effectively monitor the amount of time involved in completing assignments, and break larger tasks into smaller more manageable tasks. Parents and teachers were taught to praise and provide rewards contingent upon children's use of the organizational skills. Results of a randomized clinical trial suggest that relative to a wait-list control, those receiving the intervention received significantly better ratings of organization as reported by parents and teachers, improved academic functioning, homework, and family conflict, with large effects of the organizational skills training intervention. Compared to a contingency-based intervention which focused on parents and teachers rewarding end-goal products, organizational skills training leads to greater benefits on organizational skills as reported by parents, a difference that persisted during the follow-up assessment. Importantly, there were no differences between the organizational skills-based training and the contingency management intervention on all other outcomes. Importantly, maintenance of gains during the subsequent school year was found for both treatment groups.

There has also been considerable effort at teaching organizational skills to older youth and adolescents with ADHD. As an example, Langberg et al. (2012) adapted an organization intervention for middle-school children with ADHD, delivered within the school context by school mental health providers. The organization intervention consisted of one-to-one sessions between the child with ADHD and the school mental health professional, over the course of 16 sessions (11 weeks) that were approximately no more than 20 min/session. The organization intervention focused on training in organization of school materials, homework recording and management, and planning/time management. Parents also met with the school mental health provider for two 1-h meeting to orient themselves about the program and to help support the

use of the child's skills in the program. Compared to a wait-list control condition, the organization intervention demonstrated significant improvements at posttreatment and at 3-month follow-up in parent ratings of organization, homework, and family conflict. However, teachers did not report any differences between the two conditions. This lack of significant differences is problematic given that one would assume that teachers should be sensitive to changes in organization and homework.

The development of organizational, time management, and planning skills in youth with ADHD represents an important step in the treatment of ADHD in that these areas are impaired in many youth with ADHD and have received very little empirical attention. Although the organizational training interventions conducted by Abikoff and colleagues (2012) and Langberg et al. (2012) target the same issues, the parameters of the interventions and the study populations were quite different and offer insights into the generalizability and future directions in this line of work.

Abikoff and colleagues' intervention was a clinic-based intervention which focused on children with ADHD and documented deficits in organization, time management, and planning skills. The intervention was intensive in the sense that it was conducted during twice weekly sessions, actively involved both parents and teachers, and was conducted by well-trained, supervised clinicians. Youth who participated in this study had above-average intelligence and parents were predominantly well educated. In contrast, Langberg and colleagues' intervention was school based, focusing on children who met ADHD criteria. The intervention was developed with an eye toward generalizability and dissemination in that treatment material was simplified to the extent that session content could be delivered by a range of school mental health providers across rural and urban schools. Ultimately, materials were developed for 16, 20-min sessions conducted during the school day across 11 weeks with minimal involvement from parents and no involvement from teachers. Approximately half of the parents enrolled in the study completed college. The differences between the two studies represent the challenges found in developing interventions that balance intensity with generalizability. Future empirical studies in this area (and more broadly speaking) should keep in mind streamlining interventions to their core components (content, processes, and methods) and testing these under more broad (and routine) settings. Without this approach to treatment development and evaluation, it is difficult for a practitioner to consider the utility of implementing a particular "research-based" intervention and the appropriateness of the intervention for their populations.

Promising Interventions

Given the numerous, well-researched, and effective interventions that are available for youth with DBDs, it may be surprising that there continues to be efforts at developing new interventions. A closer look at the treatment literature, however, reveals that youth with DBDs often have insufficient response to current evidence-based interventions, limited generalizability of treatment effects to non-treated settings, lack of normalization of functioning for many treated youth, and poor longer-term maintenance of treatment gains for some youth (see Chacko et al., 2015; 2013; Kazdin, 2005; Rajwan, Chacko, & Moeller, 2012). Compounding this issue is that for the most difficult-to-treat children, there is often poor engagement to treatment (Chacko et al., 2009; Nock & Ferriter, 2005). Additionally, spurred by concerns about the development of interventions that do not fit the financial, clinical, and regulatory demands/constraints of outpatient mental health clinics in resource-poor communities (Weisz et al., 2014), there has been interest in reconceptualizing treatment delivery models for DBDs in these settings. Although a review of all the various promising treatment approaches/delivery models is beyond the scope of this chapter, we discuss Collaborative and Proactive Solutions for the treatment of DBDs, multiple family group models for the treatment of DBDs, and neurocognitive training for the treatment of ADHD.

Collaborative and Proactive Solutions

Collaborative and Proactive Solutions (CPS; Greene, 2998; 2010) is an intervention model that focuses on addressing the reciprocal adult-child coercive processes that are involved in the development of ODD by extending the focus of treatment to supporting skills in youth that contribute to the coercive process. More specifically, CPS focuses on teaching flexibility, adaptability, and problem-solving skills to both parents and children. In contrast to BPT, CPS focuses on helping parents and children learn to collaboratively and proactively solve the problems that contribute to disruptive behaviors. CPS is organized into four treatment modules:

1. Identification of underdeveloped skills and unsolved problems (e.g., completing homework, doing chores) that contribute to oppositional behavior and a discussion of the role of existing parental responses in problematic interactions
2. Prioritization—helping parents prioritize which unsolved problems will be the focus of problem-solving discussions
3. Introduction of the plan framework—helping parents understand the three potential responses to solving problems: Plan A (solving a problem from a parent-only perspective), Plan B (solving a problem collaboratively and proactively), and Plan C (setting aside the problem for future discussion)
4. Implementing Plan B—helping parents and children become proficient in using Plan B and discontinuing the use of Plan A.

The goal of treatment is to transfer the problem-solving process from one that is led by the clinician to one that is independently completed by the parent and child. Initial results of a small study of CPS compared to BPT found equivalence of treatment effects across study outcomes (Greene et al., 2004). These preliminary results warranted further empirical investigation on the benefits of CPS.

In a large randomized controlled trial of 134 youth between the ages of 7–14 with ODD, CPS was compared to BPT and a wait-list control condition (Ollendick et al., 2016). The study assessed treatment effects on disruptive behavior, impairment, remission of ODD symptoms, and ODD diagnosis at posttreatment and 6-month follow-up. Results demonstrated that CPS and BPT resulted in significant improvements relative to the wait-list control condition on all outcomes. Secondly, there were no differences between CPS and BPT on all outcomes, suggesting equivalence between the promising intervention (CPS) and a well-established intervention (BPT). Treatment gains were also largely maintained for both treatments.

Collectively, CPS is a promising approach to treating DBDs, namely, ODD, in youth. For the practitioner, it offers a different angle to work with parents and youth that focuses on the contribution of each person in the parent-child dyad in problematic interactions/behavior. While effective for many youth, the study also suggests that only 50% of youth achieve full remission of ODD, suggesting that there continues to be many youth who do not sufficiently respond to either BPT or CPS treatments. In addition, the study was a tightly controlled efficacy trial with largely middle-class, Caucasian families. As the authors note, the extent to which CPS generalizes to other practice settings and populations needs to be determined. Given that allegiance effects account for outcomes in studies, the criteria for "evidence-based" status requires independent evaluation of treatments; this appears to be a necessary step for future research on CPS.

Multiple Family Groups

Multiple family group (MFG) model was developed in response to the needs of treating youth with DBDs who present to outpatient mental health clinics in socioeconomically disadvantaged communities. While various interventions have evinced benefits for youth with DBD (e.g., BPT), there is a need to consider the extent to

which these interventions can be both readily applied in low-resource settings (e.g., community outpatient mental health clinics) with ethnic minority, multi-stressed, low-income families. The MFG service delivery model was developed to fit within the clinical, financial, and regulatory constraints of routine outpatient mental health settings in socioeconomically disadvantaged communities.

MFG takes a common elements approach (Chorpita & Daleiden, 2009; Garland, Hawley, Brookman-Frazee, & Hurlburt, 2008) by utilizing and integrating core components from the empirical literature regarding effective treatment practices for DBDs. Moreover, MFG incorporates evidence-based engagement techniques demonstrated to improve retention of families from socioeconomically disadvantaged communities in mental health services (Mckay & Bannon, 2004). These empirical literatures are translated into core skills, processes, and methods which are all framed in a strength-based perspective and are delivered in a flexible manner through a partnership model with a family partner/advocate and clinician. In addition, MFG relies on multiple generations within a family working collaboratively with other families in a group setting as a way of decreasing stigma associated with receipt of mental health services. Lastly, MFG was developed to be a parsimonious model, allowing for greater ease in training and supervising clinical staff.

Collectively, MFG was developed to increase engagement in services, provide an efficient and effective service delivery mechanism, and be readily implemented and adopted within routine publically funded outpatient mental health settings in socioeconomically disadvantaged communities. In a series of small studies conducted in socioeconomically disadvantaged community mental health clinics, youth participating in the MFG service delivery model, as compared to those participating in a services-as-usual, evinced significantly greater improvements in conduct problems, hyperactivity, impulsivity, and learning problems as well as greater retention in mental health services (McKay, Gonzalez, Quintana, Kim, & Abdul-Adil, 1999; McKay, Gonzalez, Stone, Ryland, & Kohner, 1996.; McKay, Harrison, Gonzalez, & Quintana, 2002; Stone, Mckay, & Stoops, 1996).

Recently, the MFG service delivery model was evaluated in a large randomized controlled effectiveness study (Chacko et al., 2014; Gopalan et al., 2014). Specifically, 320 youth and their families who presented to one of 13 outpatient mental health clinics serving youth and their families from socioeconomically disadvantaged communities were assigned to either participate in the MFG service delivery model or to a services-as-usual (SAU) comparison group. Results suggested the MFG model resulted in significantly improved oppositional behavior and social competence compared to the services-as-usual group. Interestingly, families who participated in the MFG required substantially less concurrent services (e.g., medication, individual therapy), suggesting that the MFG model can result in cost-savings. Additionally, treatment effects were maintained at 6-month follow-up.

The results of this large trial suggest that MFG can be delivered under routine community-based outpatient clinic settings, by existing clinical staff, treating clinic-referred youth and their families from disadvantaged communities. However, it is important to note that only 16% of youth participating in MFG achieved clinically significant improvement in ODD behaviors. This is a sobering finding but not altogether surprising. As the authors note (Chacko et al., 2014), the sample included families who were high-risk factors for poor treatment outcome (e.g., the average Beck Depression Inventory score for parents was in the moderate range; 75% of families received publically funded health insurance). While MFG represents a good start for some youth, many youth (and their caregivers) will require ongoing mental health services.

Neurocognitive Interventions

Neurocognitive interventions have received a considerable amount of interest over the past decade, particularly for the treatment of ADHD in youth. This is not altogether surprising given

the increased central role of neurocognitive deficits/executive functioning in the pathophysiology of ADHD (Halperin & Shultz, 2006). As such, ADHD has been reclassified under the rubric of neurodevelopmental disorders in the DSM-5. Efforts to develop intervention to target neurocognitive factors/executive functioning have largely been based on the lack of generalization and sustained effects of behavioral intervention and stimulant medications as well as limited effects of these interventions on key domains of impairments (e.g., academic achievement). As discussed by Chacko, Koffler, and Jarrett (2014), a potential reason for this lack of pronounced enduring effects of these interventions is that there is no compelling data suggesting that these interventions affect underlying neurocognitive/executive functions that are associated with ADHD symptoms and associated impairments. Efforts at developing treatments that more directly target neurocognitive/executive functions may result in improving ADHD symptoms but, more importantly, the impairments associated with ADHD symptoms.

To address the limitations of current ADHD interventions, researchers have begun to explore two neurocognitive treatment approaches to improve attention and executive functioning (EF) deficits. The first, facilitative intervention training (FIT), was presented in the early 2000s. The purpose of the intervention was to promote the development of attention and EFs. FIT often uses computerized (i.e., automated) training exercises to strengthen and enhance EF deficits. The primary objective of these computerized programs is that EF deficits can be strengthened over time by repetition, feedback, and practice, which in turn will transfer to other activities or abilities that depend on these executive networks (Klingberg, 2010). In other words, computerized training will produce neural changes leading to an increase in learning and development of new cognitive skills.

Current computerized FIT programs generally target either attentional, short-term memory (STM) (i.e., short-term storage and rehearsal of phonological or visuospatial subsystems) or mixed executive functions (EF) (i.e., short-term memory and behavioral inhibition). These programs are implemented either at home, school, or a clinic/research facility. The software program adjusts the level of difficulty to match the participant's performance on a trial-by-trial basis. For instance, as the participant's performance improves, task difficulty increases resulting in increased WM span.

There have been numerous studies evaluating the effects of FIT for youth with ADHD. Collectively, the data has been quite sobering. Across several meta-analytic reviews (Cortese et al., 2015; Sonuga-Barke, Bradeis, Holtmann, & Cortese, 2014; Rapport, Orban, Kofler, & Friedman, 2013), the data suggests that FIT interventions result in some improvements in ADHD symptoms as rated by reporters who were likely not blinded to treatment status (e.g., parents). However, there is no evidence that FIT results in significant improvements in ADHD symptoms or associated impairments in youth with ADHD when blind ratings are used to evaluate outcomes. As such, there appears to be no compelling data suggesting that FIT interventions should be considered appropriate interventions for youth with ADHD.

Given the state of the science, why then would we categorize FIT interventions as "promising"? There is data to suggest that FIT interventions have the potential to affect some aspects of EF. Rapport et al. (2013) found that FIT interventions, many of which targeted short-term memory (STM), did result in moderate STM improvement following training. Interestingly, the meta-analytic data suggested that these improvements in STM following training endured up to 6 months after training. This was suggested by the authors as "proof of concept" that interventions that target an EF can indeed affect the targeted outcome in a significant and lasting way. Unfortunately, STM is not a central deficit in ADHD (Willcutt, Doyle, Nigg, Faraone, & Pennington, 2005) and not related to the functional impairments associated with ADHD (Rapport et al., 2013). Future research should consider methods to better target EFs that are centrally involved in ADHD (e.g., working memory; Rapport et al., 2001) and more pur-

posefully plan for transfer effects from FIT interventions. Moreover, as we have suggested (Chacko et al., 2014) and discuss below, combining FIT interventions with skills-based interventions targeting specific functional outcomes may be a fruitful line of investigation.

The second treatment, neuro-/biofeedback, was first investigated in the 1930s to teach self-regulation. Various approaches including targeting sensory-motor rhythm, quantitative EEG, contingent negative variation, and slow cortical potentials have been proposed for the treatment of ADHD. In general, neuro-/biofeedback approaches require the patient to monitor various brain activity (e.g., through visual information), and feedback is provided when brain activity is outside of the target range, allowing the patient to adjust and self-regulate physiological activity and effect behavior (Hoffman, 2014; Lofthouse et al., 2011).

The treatment literature on neurofeedback has a long history. Initially, studies of neurofeedback suggested moderate to large effects on ADHD symptoms; however, these studies were less rigorous (e.g., nonrandomization to treatment, unreliable or subjective outcome measures). Over the past few years, there has been a significant increase in more well-designed rigorous evaluation of neurofeedback. Similar to the FIT literature, beneficial effects on ADHD symptoms have been reported in studies using reporters who were likely not blind to treatment condition; but effects are inconsistently found in studies with raters more likely blind to treatment condition (Lansbergen et al., 2011; Maurizio et al., 2014; Steiner, Sheldrick, Gotthelf, & Perrin, 2011; Steiner, Frenette, Rene, Brennan, & Perrin, 2014a; van Dongen-Boomsma et al., 2013). There are few studies of longer-term stability of treatment effects following neurofeedback, but these studies suggest some modest sustained effects (Steiner, Frenette, Rene, Brennan, & Perrin, 2014b; Meisel et al., 2013). Some, but not all (Orgim & Hestad, 2013), studies have found that neurofeedback is as effective as stimulant medication on ADHD symptoms (Duric, Assmus, Gundersen, & Elgen, 2012; Meisel, Servera, Garcia-Banda, Cardo, & Moreno, 2014). Importantly, for studies suggesting equivalence between neurofeedback and medication, medication was not optimally titrated for all study participants. As such, these studies do not compare neurofeedback to optimally prescribed medication. Collectively, neurofeedback appears to be a promising intervention for the treatment of ADHD (see similar conclusion by Evans et al., 2013) but will require further, well-controlled studies in comparison to alternative, well-implemented first-line pharmacological (i.e., stimulant medication) and non-pharmacological (i.e., behavioral treatment) interventions to determine short- and longer-term benefits of this treatment approach.

Future Directions

The literature on the treatment of DBDs is robust. There are clearly several psychosocial approaches that are effective for treating DBDs in general, as well as interventions that address ODD, CD, and ADHD specifically. Although significant, it is also clear that our most robust psychosocial interventions do not often result in normalization of functioning for many youth. As such, continued exportation into effective interventions for DBDs is necessary, particularly those that can be readily applied in routine clinical practice. We briefly close with a discussion of future directions in the treatment of DBDs in youth.

Although the DBDs have long been classified in the DSM, it is well recognized that the DBDs generally, and more specifically, ODD, CD, and ADHD, likely represent umbrella terms that represent a group of children with distinct typologies. These distinct groups may have distinct etiologies, correlates, course, and, presumably, treatment response. As an example, callous unemotional traits have gained particular empirical interest. Callous unemotional (CU) traits in youth appear to be related to distinct correlates and course (see Frick & White, 2008 for a review). While traditional BPT appears to be a very robust intervention in general for youth with DBDs, BPT combined with medication may be warranted for youth exhibiting conduct problems

and CU traits, as these youth do not respond as well to traditional BPT (Kimonis, Bagner, Linaris, Blake, & Rodriguez, 2014). Interestingly, Miller et., (2013) found that a more nuanced approach to behavioral interventions focusing on reward techniques and minimizing punishment techniques resulted in improved negative behavior (e.g., aggression) in this population. Similar findings were observed in BPT. Sanders and colleagues (2007) found that BPT was less effective for youth with DBD with CU traits. However, when effects were analyzed across the course of BPT, youth with CU traits responded equally well to positive aspects of BPT but not to punishment techniques (e.g., time-out). Importantly, CU traits are not the only subgroup that is being identified with the broader DBD construct—studies have begun to look at classes of ODD (Althoff et al., 2014) and ADHD (Campbell, Halperin, & Sonuga-Barke, 2014). Identifying subgroups of youth within the broader DBD classification may lead to better tailoring of current treatment (as is the case with callous unemotional traits in youth with CD) as well as offer an opportunity to develop novel approaches that are better tailored to treat these distinct groups.

Abikoff et al. (2013) offer a unique approach to allocating treatment. Rather than focusing on a diagnosis (e.g., ADHD), these investigators targeted youth with a diagnosis (i.e., ADHD) who had a deficit in a skill (i.e., organization, time management, and planning) which was the target for intervention (i.e., organizational skills training). This "needs-based" approach seems like common sense—treat the problem not the diagnosis. In fact, it is very much aligned to traditional behavior therapy principles. Alas, the movement toward evidence-based treatments for specific diagnosis has moved much of the empirical investigation into treatments away from inclusion of youth into studies based on skills deficits (i.e., "a need"), toward inclusion of children based on diagnostic status. Given the heterogeneity of children within current DSM classifications of ODD, CD, and ADHD (as discussed above), inclusion of youth based on a diagnosis may attenuate treatment response. As an example, a review of the literature of traditional social skills interventions suggests that there is no clear support for these interventions for youth with ADHD. A "needs"-based perspective may shed some light into these findings.

First, not all youth with ADHD have social skills deficits (Hoza, 2007a, b). Theoretically, children with ADHD and significant social skills deficits would likely benefit most from treatment targeting social skills. Specifically, those with a higher level of social impairment at baseline should have more opportunity to improve from a treatment specifically targeting social skills than those who do not have significant baseline social impairment. To our knowledge, only three studies to date (Fenstermacher et al., 2006; Pfiffner & McBurnett, 1997; Sheridan et al., 1996) have explicitly used objectively measured social skills deficits as a criterion for inclusion. These studies suggest, in contrast to the broader literature, that social skills interventions may be a useful for youth with ADHD with a social skills deficit. This needs-based perspective should be further studied to better understand the effects of current interventions as well as when developing new intervention approaches for treating various skills deficits in youth with DBDs.

Combined treatment approaches have been a focus of treatment for DBDs, often including the combination of skills-based intervention (e.g., BPT plus PSST) or psychosocial treatment with medication. There is a growing appreciation for the role of neurocognitive deficits in the pathophysiology and/or maintenance of a host of psychiatric conditions (e.g., schizophrenia, Green & Nuechterlein, 2004; anxiety, Bar-Haim et al., 2007; depression, Siegle et al., 2007), including ADHD (Willcutt et al., 2005). While the neurocognitive treatment literature, particularly FIT interventions, has not been clearly shown to improve symptoms or functioning in youth with ADHD, data suggests that aspects of executive functioning (i.e., short-term memory) can be improved following FIT treatments (Rapport et al., 2013). This proof of concept is important—targeted, adaptive training can have an effect on executive functioning in youth with ADHD. If future interventions can better target more central executive functioning deficits (e.g.,

working memory; Rapport et al., 2001), this may lead to more pronounced effects on outcomes. While this may be important, simply improving executive functions may be insufficient in maximizing outcomes for youth with ADHD. As we have articulated in greater detail elsewhere (Chacko et al., 2014), we view the relation between improved neurocognitive functioning and functional outcomes in ADHD as akin to the relation between corrective lenses and reading: glasses allow children with farsightedness to see the printed words and benefit from classroom instruction, but the glasses themselves do not teach children, who have acquired deficits in reading over time, to read. As such, improving executive functions is expected to result in improved *potential* due to the improved cortical foundation, but changing the trajectory of ADHD-related interpersonal difficulties and academic underachievement will likely require targeted, individualized, skill-focused interventions to remediate specific skills that were not mastered previously due to neurocognitive limitations. We believe that carefully combining neurocognitive interventions that target executive functions that are related to a particular outcome (e.g., reading) along with interventions to remedy skills deficits in this area (phonologic awareness training for reading), one can maximize functional improvements (e.g., reading) for youth with ADHD. Importantly, this combined neurocognitive plus skills-based approach has garnered support in other psychiatric conditions (schizophrenia; Bowie et al., 2012; anxiety, Shechner et al., 2013; depression, Richey et al., 2013), suggesting that this is a promising area to pursue.

Randomized controlled trials of interventions often focus on the head-to-head comparison of interventions with a fixed endpoint. Given the variability in response to evidence-based treatment and the variable nature in the clinical presentation of youth with DBDs (e.g., comorbidities, skills deficits, etc.), future intervention research should also focus on how best to combine, sequence, and modify treatment(s) over time based on ongoing response to treatment. This adaptive treatment approach (Collins et al., 2004) reflects the nature of clinical practice—providers must consider which treatment(s) to use for a child, determine if there is a need to alter treatment plans based on how well goals are being achieved, and what is the appropriate subsequent treatment(s) to utilize given previous response to treatment(s). This approach offers an opportunity to address clinically meaningful questions. As an example, Pelham and colleagues (2013) are evaluating an adaptive treatment approach for youth with ADHD where stimulant medication and behavioral intervention are sequenced, switched, and/or augmented based on the effects of treatment progress monitoring of functional impairments at home and school over time. This study offers the opportunity to address questions such as which treatment should be initialized when starting a treatment regimen for youth with ADHD and does this vary depending on characteristics and preferences of the youth/family? Unfortunately, there is a scarcity of research on adaptive treatment designs for treating DBDs in youth. The field, however, is primed for such approaches given the number of evidence-based interventions currently available to address various functional impairments and skills deficits in youth with DBDs. This remains an important area of future investigation.

Lastly, increasing attention has been paid to the generalizability of evidence-based treatments to routine practice settings (Weisz et al., 2014). There remains a significant gap between what we know is effective in research and the use of these interventions in practice. The aforementioned future directions offer interesting and potentially valuable approaches to better treating youth with DBDs. However, these various approaches must take into account the regulatory, financial, and clinical realties of routine practice. Conceptual models that can help guide the intervention development process has been articulated (e.g., Hoagwood, Burns, & Weisz, 2002; Weisz, 2004) and should be considered as an integral part of the development of novel interventions, combined approaches, or treatment algorithms. Ultimately, intervention development without an eye toward the people being served, the providers delivering treatment, and the final practice context where the intervention is to be deployed

(e.g., schools, outpatient mental health clinics) will only result in further widening the research to practice gap.

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Cognitive and Behavioral Interventions to Improve Social Skills

33

Mi Na Park, Ruth Ellingsen, and Elizabeth A. Laugeson

Introduction

Impairment in social functioning is a hallmark feature of many childhood psychiatric disorders. Youth who commonly present for social skills treatment include those diagnosed with autism spectrum disorder (ASD; Carter, Davis, Klin, & Volkmar, 2005; White, Keonig, & Scahill, 2007), learning disorders and intellectual disability (Forness & Kavale 1996; Gresham, Sugai, & Horner, 2001; Guralnick, 2005), and fetal alcohol spectrum disorder (Bishop, Gahagan, & Lord, 2007; Thomas, Kelly, Mattson, & Riley, 1998). In addition, those with internalizing disorders, such as anxiety and depression (Segrin, 2000; Spence, Donovan, & Brechman-Toussaint, 1999), and externalizing disorders, such as attention-deficit/hyperactivity disorder (ADHD), emotional and behavioral problems, and conduct disorders (Kavale, Mathur, Forness, Rutherford, & Quinn, 1997; Pelham & Fabiano, 2008; Webster-Stratton, Reid, & Hammond, 2001), also often require significant assistance in their peer interactions and relationships. However, the social challenges ascribed to each group may be unique. For instance, children with emotional and behavioral problems largely struggle to adapt to demands and social expectations, while children with ADHD often struggle with inhibition of socially inappropriate responses and distractibility during social problem-solving (Carter & Lunsford, 2005; Spence, 2003). Ultimately, the consequences of these social deficits surpass diagnostic categories and often result in peer rejection, peer isolation, and lack of meaningful, reciprocal friendships.

The impact of these social difficulties is pervasive, extending beyond strained peer interactions and poor quality of friendships. Social challenges in childhood have been consistently correlated with problems across other domains of functioning, including psychological, behavioral, academic, and adaptive living. Moreover, youth with social difficulties are also at higher risk for peer rejection, loneliness and social dissatisfaction, anxiety, depression, academic problems, early withdrawal from school, increased aggression, delinquency, and even legal troubles (Coie, Terry, Lenox, Lochman, & Hyman, 1995; DeRosier, Kupersmidt, & Patterson, 1994; Elliott, Malecki, & Demaray, 2001; Howlin & Goode, 1998; Hymel, Rubin, Rowden, & LeMare, 1990; Whitehouse, Durkin, Jaquet, & Ziatas, 2009). Parents of these youth often report the onset of peer difficulties in early childhood, with social problems persisting and magnifying with development. A notable increase in social

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problems emerges in adolescence when social demands and expectations magnify alongside with greater sensitivity to and awareness of social relationships (Nelson, Leibenluft, McClure, & Pine, 2005). Without the appropriate intervention, these problems escalate and persist into adulthood.

Conversely, one of the strongest predictors of overall well-being in adulthood is peer acceptance during childhood (Parker & Asher, 1987). Research suggests that socially adept youth with positive peer interactions and healthy friendships are less likely to develop mental health problems such as anxiety and depression, describe greater support and satisfaction in their relationships, and endorse greater academic engagement and achievement (Armsden & Greenberg, 1987; Deković, Buist, & Reitz, 2004; Lansford, Criss, Pettit, Dodge, & Bates, 2003; Malecki & Elliott, 2002; Ryan, Stiller, & Lynch, 1994). Moreover, even having one or two close friends may improve self-esteem and decrease the impact of daily life stressors (Buhrmester, 1990; Miller & Ingham, 1976). As such, social skills training programs have become critical components of comprehensive treatment plans for most youth presenting with social, behavioral, and neurocognitive concerns. Not only do these social interventions strive to improve immediate psychosocial functioning, they also aim to prevent or ameliorate future maladjustment (Spence, 2003).

Cognitive and Behavioral Social Skills Interventions

Most neurotypically developing youth often learn basic rules of social etiquette through a combination of informal observation of their peers and more formal instruction from parents in their day-to-day interactions (Bandura, Ross, & Ross, 1963; Rubin & Sloman, 1984). However, there is a large minority of youth that require additional, more structured support and assistance. This need for more formalized instruction is often met through social skills training, which teaches specific social skills using cognitive, behavioral, and/or social learning strategies. Broadly speaking, cognitive and behavioral social skills interventions aim to decode and simplify developmentally appropriate yet complex social behaviors into concrete steps and strategies. This simplification enhances the child's comprehension of the skill and increases the likelihood of correct application and use of the skill in their everyday social lives (Cappadocia & Weiss, 2011).

Given that social skills interventions continue to grow in popularity, it is important to examine its literature base. A review of the existing literature shows that outcomes of many social skills interventions are variable and inconclusive. While some meta-analytic studies show that social skills interventions are an effective form of treatment for specific populations (e.g., emotional and behavioral disturbances; Cook et al., 2008; Gresham, 2015), other meta-analytic studies have shown that social skills interventions by and large only lead to modest outcomes that do not meaningfully persist beyond the conclusion of treatment (Ang & Hughes, 2002; Cook et al., 2008; Kavale & Mostert, 2004; Maag, 2005, 2006; Quin, Kavale, Mathur, Rutherford, & Forness, 1999; Spence, 2003). Delivery of treatment in un-naturalistic treatment settings, failure to teach ecologically and socially valid social skills, lack of consideration for the unique social needs and learning styles of different groups of children and adolescents, minimal or lack of active parent involvement, and failure to generalization skills to natural social settings are a few factors hypothesized to attenuate the quality and extent of meaningful, clinically significant treatment gains (Frankel, Myatt, Cantwell, & Feinberg, 1997; Gresham, 1998; Maag, 2006).

Another reason that may account for some of the variability in outcomes is the expansive breadth of social skills interventions. The wide scope of social skills program may be reflective of the distinct social needs and unique set of broader challenges (e.g., cognitive functioning, language abilities, neurocognitive impairments, etc.) exhibited by targeted groups (e.g., ASD, ADHD, fetal alcohol spectrum disorder, etc.) (Spence, 2003). Programs vary in the types of skills they target, some focusing solely on one

skill (e.g., conversation skills) and others teaching a broader set of skills (e.g., friendship skills) (Kaat & Lecavalier, 2014). Specific skills or areas of focus range from theory of mind (Ozonoff & Miller, 1995) to social cognition or social perception (i.e., using conventional social knowledge to respond to social situations; Crooke, Hendrix, & Rachman, 2008) to interpersonal problem-solving (i.e., solving common social problems; Shure & Spivack, 1980).

Findings from several meta-analyses examining the effectiveness of social skills interventions suggest that multicomponent programs that incorporate strategies based on cognitive, behavioral, and social learning perspectives may be most beneficial in producing meaningful outcomes. Similarly, a social skills program that simultaneously addresses other developmental factors may be even more effective in enhancing social competence as these factors may potentially compete with a child's capacity to benefit from treatment (Gresham, 1998; Spence, 2003). Spence (2003) argued that social skills knowledge is not exclusively indicative of social competence. Other cognitive, affective, and environmental factors that transactionally impact social behavior and functioning must also be addressed. For instance, considering whether a child may have maladaptive thinking patterns (cognitive), difficulty regulating their anger or anxiety (affective), and/or has access to sufficient and appropriate social opportunities (environmental) and concurrently addressing these issues may maximize response to treatment.

In spite of the mixed results regarding the extent to which social skills instruction may be helpful, it is clear that the benefits of social skills instruction are optimized and most appropriate when integrated into a comprehensive treatment plan rather than a stand-alone intervention (Spence, 2003). For instance, social skills interventions have been shown to be effective for specific childhood disorders such as social phobia and learning disabilities (Forness & Kavale, 1996; Spence, Donovan, & Brechman-Toussaint, 2000) when provided as part of a larger treatment program. This is a likely a reflection of the broader challenges presented by specific psychiatric disorders that impact social functioning, as well as the high rates of psychiatric comorbidity among disorders that present for social skills instruction. Other studies have shown that social skills instruction alone is insufficient in improving social outcomes; it must be provided within the broader context of other evidence-based strategies, such as parent training, behavior management, and self-regulation (Spence, 2003). Similarly, Frankel et al., (1997) found that children with ADHD benefited most from an integrated approach that included parent training, social skills instruction, and medication management.

This chapter will focus on intervention strategies that aim to teach children and adolescents social skills that are fundamental to positive peer interactions and satisfying, reciprocal friendships through cognitive and behavioral treatment modalities (Gresham, 1998; Maag, 2005). By learning fundamental social skills, the expectation is for the child or adolescent to flexibly apply the newly learned skills across a range of social contexts, thereby ultimately increasing their social independence and social competence (Spence, 2003). Several treatment modalities and evidence-based methods of social skills instruction are presented.

Social Skills Intervention Modalities

Cognitive and behavioral social skills instruction can be offered through various modalities, which include group-based instruction, adult-mediated programs, parent-assisted interventions, peer-mediated programs, and school-based or teacher-facilitated interventions.

Group-Based Instruction

Group instruction is an intuitive method for social skills training, as it offers children and adolescents the opportunity to interact with and practice newly learned social skills with their peers. When providing group-based social skills training, several elements should be considered.

First, group facilitators should have a shared understanding of each child's history and specific needs in order to foster group cohesion and support. Heterogeneity of the group should be assessed. For some populations, such as for children with ASD, group heterogeneity should be limited in order to aid learning and group cohesion (White, 2011). For other populations, however, such as children with oppositional behaviors or conduct problems, a heterogeneous group may be more suitable as negative behaviors are less likely to be reinforced or modeled during the group sessions (Ang & Hughes, 2002). Despite these considerations, some disruption from group members is still likely to occur. As such, a small group size (7–10 group members) is ideal.

Another important consideration relates to the gender and age range of the group members. Although gender and age of group members ought to be considered when forming groups, it may be difficult to create groups with equal gender balance (e.g., externalizing disorders and ASD are more commonly diagnosed in males than females). Whenever possible, a mixed-gender group may be helpful as it reflects the natural setting for most children and adolescents outside of the treatment setting (White, 2011).

Adult-Mediated Programs

In adult-mediated programs, social skills instruction is often delivered in the context of highly structured, one-on-one therapy sessions between child and therapist. While a therapist-driven, individual treatment model was prevalent when social skills intervention was in its infancy, it has garnered criticism largely due to a mismatch between the teaching context and the child's natural social world (i.e., lack of ecologically valid social context). That is, teaching in one-on-one treatment settings strips social meaning from the interaction, thereby diminishing potential for skill generalization (Gresham, 1998). This approach oftentimes results in context-specific responding and reduces generalization and maintenance of the newly learned skills. That is, an adult must always be present to prompt the behavior, and the child or adolescent exhibits the newly learned behavior under two restricted conditions – in the presence of that specific adult/therapist or when the reinforcer used to teach the behavior is immediately accessible (Strain, Cooke, & Apolloni, 1976).

The effectiveness of adult-mediated social skills programs may be enhanced using group-based treatment modalities, which offer a more naturalistic setting, in combination with more realistic social supports (e.g., parent assistance, peer mediation, teacher facilitation). Alternatively, this approach may be clinically indicated when initially treating youth who require a higher level of individualized attention. Youth with severe anxiety, aggressive outbursts, and significant affective lability may not have the capacity to benefit from group treatment or peer-mediated intervention initially, and their participation in such treatment modalities may be harmful and distressful to those around them. Thus, appropriateness of fit to treatment modality, whether it be group-based or individually focused, should be considered.

Parent-Assisted Interventions

Parent involvement in social skills interventions has been found to significantly improve the acquisition of social skills and treatment outcomes in several studies (Frankel et al., 1997; Laugeson, Frankel, Gantman, Dillon, & Mogil, 2012; Laugeson, Frankel, Mogil, & Dillon, 2009; Maag, 2005, 2006). By including parents in treatment, they can encourage and support their child's flexible use of newly learned skills in their natural social environments (e.g., home, school, neighborhood, etc.), thereby promoting maintenance and generalization of the skills outside of the treatment setting. Moreover, parents can play a pivotal role in identifying the ideal setting (e.g., specific place or peer group) for their child to use the newly learned skills. By helping to find an appropriate source of friends, parents may increase the likelihood that their child's attempts will be met with positive peer feedback, thereby

promoting durability and generalization of the skills (Spence, 2003). This can be a particularly transformative and corrective process for peer-rejected children and adolescents who have a long history of peer rejection despite their best efforts and intentions.

Two evidence-based programs that actively include parents in treatment with remarkable outcomes are the Children's Friendship Training program (CFT; Frankel & Myatt, 2003) for elementary-aged children and the Program for the Education and Enrichment of Relational Skills (PEERS®; Laugeson & Frankel, 2010) for adolescents with ASD. Long-term follow-up of children and adolescents who participated in CFT or PEERS® showed that these youth maintained their treatment gains 1–5 years post-intervention (Mandelberg, Frankel, Cunningham, Gorospe, & Laugeson, 2013; Mandelberg et al., 2014), strongly supporting the call for parent participation. Despite these promising findings, few social skills interventions incorporate a parent component.

Peer-Mediated Interventions

In peer-mediated interventions, other children and adolescents are used to deliver treatment (Odom & Strain, 1984). Peer-mediated approaches to social skills intervention are based on the premise that children naturally develop social skills and learn about the social world through ordinary interactions with other children their age. Odom and Strain (1984) suggested that these interventions vary in the roles assumed by the peers. Some peer-mediated interventions target direct interaction between the socially challenged youth and his/her peers by calling on peers to prompt and reinforce social behaviors, initiate social interactions with the target child, and/or be in close proximity to the target child to increase the likelihood of social interactions. On the other hand, indirect interventions do not involve direct interaction with the target youth and may consist of modification of group reinforcement contingencies (e.g., group reinforcement for cooperative and reciprocal social interactions) and the use of peers as models for appropriate behavior (Strain et al., 1976).

One of the strongest arguments for peer-mediated interventions is the greater potential for generalization as skills are modeled and practiced in naturalistic settings (Mathur & Rutherford, 1991). Peers can be invaluable in continuously prompting and reinforcing a child's social attempts across a range of social contexts (Strain et al., 1976). Consequently, peer-mediated interventions have demonstrated greater ecological validity than adult-mediated interventions (DiSalvo & Oswald, 2002).

School-Based or Teacher-Facilitated Interventions

School-based social skills interventions are often widespread, supported by local, state, and federal efforts. The impetus to address student's social competence follows a growing body of research showing that social competence is associated with psychosocial health, school performance, and academic achievement (Gresham & Elliott, 2014). Despite the popularity of school-based social skills instruction, relatively little research has been conducted to support its effectiveness. Many of these programs are led by speech and language pathologists, special education teachers, and school psychologists who develop their own programs based on a combination of existing interventions and clinical hunches rather than an evidence-based curriculum. This eclectic approach is most likely in response to limited availability of empirically supported school-based curricula. The notion that teachers can effectively teach social skills in the classroom, much like teaching math or science, is a novel approach but is slowly gaining research evidence (Laugeson, Ellingsen, Sanderson, Tucci, & Bates, 2014). The use of teachers as social skills facilitators may be a nice alternative to traditional adult-mediated social skills interventions as this modality has the capacity to reach a greater number of children in a more natural social environment.

Effective Treatment Delivery Methods for Social Skills Training

Research suggests that there are several key ingredients needed to successfully teach social skills. Effective treatment delivery methods for social skills instruction include the following cognitive and/or behavioral methods:

- Behavioral modeling and role-playing demonstrations
- Behavioral rehearsal exercises with performance feedback
- Use of social stories
- Video modeling and video self-modeling
- Use of technology and multimedia software
- Self-monitoring and self-management

Behavioral Modeling and Role-Playing Demonstrations

Behavioral modeling, or role-playing demonstrations, is a critical component in cognitive and behavioral social skills training. This method of instruction, which involves the demonstration or acting out of targeted behaviors, is particularly important in social skills training as it transforms theoretical or abstract concepts into realistic, concrete, and meaningful concepts and strategies (Spence, 2003). It is largely based on a social learning perspective, which asserts that youth naturally learn social skills from watching social models perform social behaviors in their everyday lives (Bandura et al., 1963). Behavioral modeling can be accomplished by watching a video demonstration of the targeted skill (i.e., video modeling) or by observing social coaches perform the skill. Several factors should be considered in maximizing the impact of behavioral modeling. First, to the greatest extent possible, social coaches or models should be similar to the children and adolescents (e.g., age, appearance, and background). In addition, social coaches should provide a realistic portrayal of the skill rather than a flawless, unrealistic performance and incorporate references or props that youth are likely to experience in their natural social world (Spence, 2003).

Behavioral Rehearsal with Performance Feedback

Ongoing practice of newly learned skills is vital to skill-building and generalization. This can be completed in-session through behavioral rehearsals with performance feedback from trained social coaches in order to correct behavioral errors and cognitive distortions (Blagys & Hilsenroth, 2002). Outside of the treatment setting, performance feedback can be provided by peer mentors, parents, teachers, or other trainers (Spence, 2003). Having multiple social coaches across environments to prompt the child and provide feedback is ideal as it discourages prompt dependency on a specific social coach (White, 2011).

There are several benefits to in-session behavioral rehearsal with performance feedback. For one, practice in a supportive and comfortable setting may result in the child experiencing lower anxiety and increased confidence in using the new skill in their natural social environment. Additionally, in-group practice allows the treatment team to assess the child's comprehension and implementation of the skill and provide corrective feedback when necessary, thereby increasing the potential for positive responses from peers when used outside of the treatment setting. When providing feedback, it is essential to balance constructive feedback with ongoing praise for the child's efforts in using the newly learned skills (Spence, 2003).

Social Stories

Another commonly used cognitive method for teaching social skills involves the use of social stories. This technique uses short stories to describe common social situations, focusing on relevant social cues, perspectives, and common social responses (Gray & Garand, 1993; Gray, 1998, 2000). The stories are most often written from the perspective of the child (first-person perspective) in order to aid the child's ability to relate with the social situation presented in the book. The aim of social stories is not necessarily to modify the child's behavior, but, rather, to

improve the child's understanding of social events and expectations (e.g., describe why the social situation is occurring, identify the relevant people/characters, depict an appropriate social response). Through this descriptive approach, the goal is to increase awareness of social situations or events that may then eventually generalize to the child's use of appropriate social responses (Gray & Garand, 1993; Gray, 1998, 2000). While this technique is commonplace in school settings and preliminary research findings are encouraging, additional research is warranted. Initial research suggests that social stories may be most effectively used as part of a multicomponent intervention that incorporates other evidence-based strategies such as video modeling, prompting, and reinforcement (Scattone, 2007).

Video Modeling and Video Self-modeling

A form of observational learning, video modeling, involves watching a video demonstration of a desired social behavior and then imitating the observed behavior (Grant & Evans, 1994). For instance, in teaching the skills related to peer entry, a child might watch a video of a child successfully entering a conversation among a group of children and then practice the skill by imitating the child portrayed in the video. Likewise, video self-modeling consists of the child watching him- or herself successfully perform a desired social behavior on video, followed by imitation of the targeted behavior (Dowrick, 1999). By recording and re-watching their own performance, treatment staff and social coaches can more readily identify specific aspects of the skill that may require additional refinement and corrective feedback.

The potential for video modeling in enhancing social competence is encouraging. Charlop-Christy, Le, and Freeman (2000) compared the effects of video modeling to in vivo modeling (therapist-led role-play demonstrations) in teaching children with autism spectrum disorder a variety of skills (i.e., conversation skills, play skills, and self-help skills). Skills that were taught via video modeling were acquired more rapidly and led to greater generalizations than skills taught using in vivo modeling. The power of video modeling may be attributed to its novelty as a mechanism for learning and association with recreation and entertainment, all of which promote motivation and attention during learning (Dowrick, 1986). One of the drawbacks of video modeling, however, is the amount of effort dedicated to video preparation, which exponentially grows for each specific skill that needs to be targeted (Scattone, 2007).

Technology and Multimedia Software

The use of technology in the delivery of social skills treatment is an expanding area of study that continuously grows with ongoing advances in technology. Video modeling and video self-modeling, virtual reality, and audio scripts are some of the common technology and multimedia components that have been integrated into social skills treatment. These approaches are becoming more and more popular given their capacity to deliver treatment to a large number of youth in a resource and fiscally efficient manner, consistently deliver treatment at high fidelity, and present repeated practice opportunities without personnel fatigue (Cromby, Standen, & Brown, 1996; Reed, Hyman, & Hirst, 2011).

One of the most appealing applications of technology is virtual reality or virtual environments. Through the real-world simulations offered by virtual reality, the child or adolescent may steadily acclimate to social situations that may have been too emotionally charged (Parsons & Mitchell, 2002). In addition, virtual environments may promote greater flexibility in and generalization of the newly learned skills as the child can proceed through a set of simulations that systematically increase in complexity based on the child's progress.

While alluring, not all service providers have the resources to incorporate these technology-based approaches. Platforms such as virtual environments necessitate a high level of training to develop and fine-tune the program in line with skill acquisition and sufficient knowledge of technology is needed by the treatment team in order to train children and adolescents in its use (Cumming, 2010).

Self-monitoring and Self-management

Several studies have demonstrated self-monitoring, or self-management, to be an efficacious intervention strategy in teaching social skills. Self-control approaches typically consist of self-assessment (observing one's own behavior), self-evaluation (evaluating one's behavior in relation to a goal or target behavior), self-recording (recoding one's demonstration of the target behavior using tokens, wrist counters, etc.), and self-reinforcement (rewarding oneself upon meeting the goal) (Bandura, 1976). This strategy has been frequently used with good outcomes for teaching social skills (e.g., social initiations and play skills) to children with ASD (Koegel, Koegel, Hurley, & Frea, 1992; Morrison, Kamps, Garcia, & Parker, 2001; Stahmer & Schreibman, 1992). The use of self-monitoring and self-management strategies is especially appealing because it can help children and adolescents become more socially independent and less reliant on ongoing assistance from parents, educators, and professionals (Hume, Loftin, & Lantz, 2009).

Other Considerations for Cognitive and Behavioral Social Skills Training

Other key features thought to enhance and track treatment outcome for children and adolescents receiving cognitive and behavioral social skills training include:

- Using evidence-based treatment manuals
- Providing sufficient duration of treatment
- Implementing didactic instruction
- Teaching ecologically and socially valid social skills
- Using in vivo socialization homework assignments

Evidence-Based Treatment Manuals

The use of evidence-based treatment manuals may help to ensure that children and adolescents in community treatment settings achieve comparable treatment gains as research participants upon which the research is based. Although actual delivery of treatment may still vary, using treatment manuals may help to standardize interventions (Smith et al., 2007). Efforts toward treatment fidelity or adherence to original guidelines for delivering the intervention may be particularly important for interventions delivered by parents or less experienced or credentialed mental health professionals (Matson, 2007). These efforts may consist of observation, regular meeting with and training of treatment staff, ratings of treatment fidelity during sessions, and use of structured manuals and guides (Spence, 2003).

The goals of treatment fidelity, however, must be balanced with the reality of community settings. For example, ideally clinicians would receive training on a particular intervention until reliability in their delivery of the intervention was reached. However, this may not be feasible due to the extensive time and financial costs involved in training and ongoing supervision of clinicians. A more realistic option for maintaining treatment fidelity may be to have an assistant or coach monitor a checklist of targeted points to be covered during delivery of treatment sessions to ensure that all elements are covered. As long as treatment fidelity is maintained, the use of evidence-based treatment manuals is helpful toward effective dissemination and replication of empirically supported treatments in community settings.

Duration of Intervention

Some time-limited cognitive and behavioral social skills training interventions involve instruction over a short period of time, such as a few of weeks to several months. However, social skills instruction in the community is most often provided on an ongoing basis with no predetermined time line or end point; yet, the efficacy of this approach has not been thoroughly examined. Although the specific advantages and disadvantages of time-limited social skills interventions are unclear, it is likely that this model will become more widespread and mandated due to

the constraints of managed healthcare. Whatever the case may be, duration of intervention is an important consideration to make when designing the format of a social skills intervention and aims of the program, and it should be based upon the needs of the target youth population. When considering duration and frequency of intervention within the school setting, Spence (2003) recommends provision of social skills instruction throughout the year in a stepwise fashion, much in the way other core academic subjects are taught. Other recommendations include booster sessions or maintenance programs (Spence, 2003).

Didactic Instruction

The use of structured lesson plans to teach social skills using concrete rules and steps of social behavior is also key to the successful implementation of an effective cognitive and behavioral social skills program (Laugeson & Frankel, 2010; Laugeson, 2013; Laugeson, 2014). Structured lesson plans ensure that a core set of skills will be taught. Many community-based social skills programs approach social skills instruction through "process groups" in which children and adolescents are asked to recount their week, while therapists and other group members attempt to troubleshoot potential problems and brainstorm more socially constructive responses. The advantage of this process-based approach is unknown, and the risk of possibly failing to teach a core set of skills imperative to function adaptively in the social world may outweigh any benefits. The use of structured didactic lessons is recommended to ensure that a predetermined fundamental set of skills is learned.

Additionally, when providing social skills instruction to children and adolescents, it is important to consider the unique cognitive style of the targeted children and adolescents. For example, youth with ASD typically think in very concrete and literal terms. Therefore, didactic lessons should be presented using concrete rules and steps of social etiquette, while use of metaphors, analogies, and other forms of figurative language, which those with ASD often struggle to comprehend, should be avoided (Kerbel & Grunwell, 1998; Starr, Szatmari, Bryson, & Zwaigenbaum, 2003).

The use of structured didactic instruction should not be limited to children and adolescents and should extend to parent sessions. Frankel et al., (1997) found improvements in skill generalization for children with oppositional defiant disorder following participation in a social skills intervention, which was in contrast to previous findings. The authors hypothesized that the didactic lessons provided parents with specific skills to increase prosocial behaviors and decrease disruptive behaviors and that the structured session format was key in increasing completion of socialization homework assignments and generalization of skills.

Teaching Ecologically and Socially Valid Social Skills

Ecologically valid social skills are those behaviors that are naturally exhibited by socially accepted youth in a given social context. Relatedly, socially valid social skills are those behaviors that will meaningfully improve the quality of life for the target youth. Unfortunately, many social skill programs teach neither ecologically valid nor socially valid social skills (Maag, 2005; Spence, 2003). Rather, social skills instruction includes social behaviors and rules of social etiquette deemed appropriate by adults, clinicians, or researchers, rather than those desired by the target youth or rules established by the dominant peer group. This is problematic, as youth will disengage from treatment and, more importantly, continue to experience social difficulty.

A good example of this involves peer entry, which is how people enter group conversations. What are most children and adolescents told to do to enter conversations and meeting new people? They are often encouraged to approach the group and introduce themselves or go up and say, "Hi." Unfortunately, this advice is not ecologically valid. Instead, research suggests that children and adolescents typically meet one another by

entering group conversations focused on common interests. First, they discreetly watch and listen to the conversation. A prop like a mobile phone or gaming device can be helpful in doing this unobtrusively so they will not appear to be eavesdropping. While listening, they are identifying the topic of conversation and assessing if they can meaningfully contribute to the conversation through common interests. Once they have decided to join, they wait for a pause in the conversation, so as not to interrupt the conversation, and move a little closer (approximately an arm's length away) in order to show interest in the group. Then they join by making a comment, asking a question, or giving a compliment related to the topic (Laugeson & Frankel, 2010). Teaching ecologically valid social skills is a far more effective method of social skills instruction, leading to greater peer acceptance and social competence.

In Vivo Socialization Homework Assignments

One of the common criticisms of social skills training programs is that the skills taught do not generalize to other environments or maintain once treatment concludes. This can be addressed through the provision of socialization homework or practice assignments that are completed in between sessions, a common cognitive and behavioral technique used to enhance generalization. For example, in the case of conversational peer entry, children might be assigned to practice the steps for peer entry (i.e., watch/listen, wait, join) at school or at an extracurricular activity. Providing socialization homework assignments alone is insufficient. A portion of each session (preferably at the beginning) should be dedicated to reviewing attempted or completed homework assignments, as well as troubleshooting any obstacles or issues that may have arisen. To increase homework compliance and boost the likelihood of successful practice, the format in which the assignments are provided should also be considered. Outlining the assignments on handouts or "home-task cards" with explicit details (e.g., location, time, peers with whom to practice) allows the treatment staff to proactively address any potential issues or barriers to homework completion (Spence, 2003).

Assessing Treatment Outcome

Refined assessment procedures before and after treatment are essential to understanding the true benefits of cognitive and behavioral social skills interventions, as well as identifying who is most likely to benefit from targeted treatment. For example, irrespective of diagnoses, most social skills programs are broadly targeted for children and adolescents with social deficits. However, researchers have identified two distinct types of social deficits – acquisition deficits and performance deficits. It is important to know if a child is experiencing social challenges primarily due to acquisition or performance deficits, as this will dictate the proper type of social skills intervention (Gresham, 1981; Gresham, 2015). Youth with acquisition deficits would be most helped by programs that focus on teaching specific skills, whereas youth with performance deficits would benefit most from programs that focus on modification of reinforcement contingencies to promote the performance of skills and appropriate social behavior. For those youth struggling with both types of social deficits, a combination of these methods would be better suited. Assessing treatment outcome not only provides useful information about who is most likely to benefit from treatment, but allows us to make informed decisions in developing targeted treatments for youth with diverse social challenges.

Whatever the presenting challenges of children and adolescents struggling with social skills deficits, assessment of treatment outcome is a critical ingredient to ensuring that social skills training is effective and beneficial.

Conclusion

Deficits in social skills cross several diagnostic categories, affecting the lives of children and adolescents at home, at school, and in their community. Without appropriate intervention, these social

challenges are likely to worsen and exacerbate with development, diminishing psychosocial and adaptive well-being into adulthood. While ongoing research is needed in better assessing the effectiveness of social skills interventions and its impact on social competence and well-being, there do appear to be effective cognitive and behavioral treatment components and strategies that enhance treatment benefit. These treatment components include behavioral modeling or role-playing demonstrations, behavioral rehearsal exercises with performance feedback, social stories or scripts, video modeling or video self-modeling, the use of technology and multimedia software, and self-monitoring or self-management.

Other important considerations in maximizing the potential benefit of cognitive and behavioral social skills interventions include using evidence-based treatment manuals with didactic instruction of ecologically and socially valid social skills, using in vivo socialization homework assignments to assist in generalization and maintenance of newly learned skills, providing sufficient duration and frequency of treatment appropriate to the target population, and tracking treatment outcomes in order to understand the utility of social skills treatment.

In conclusion, as the empirical support for cognitive and behavioral social skills training for children and adolescents continues to grow, the need for evidence-based treatments for specific psychiatric populations is warranted. The benefits of explicit programming for generalization and use of comprehensive assessment procedures to better characterize and treat the target population should be specifically examined to improve the social outcomes of children and adolescents. The demand for effective social skills interventions specific to diagnostic groups, yet sensitive and flexible enough to meet their complex and broader needs, has never been greater.

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