CHAPTER 21

Intellectual Disabilities

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In DSM-5 and in several DSM editions, intellectual disability has been the first disorder described. In spite of this prominent position, however, the study of children and adults with intellectual disabilities generally lags behind interest in other psychiatric conditions. Yet within psychiatry there has long been a sense that interest in intellectual disabilities is increasing. Ninety-one years ago—in 1927—Howard W. Potter described intellectual disabilities as the neglected child of psychiatry, which he considered the Cinderella of medicine. Like Cinderella, Potter noted, intellectual disabilities would soon take its rightful place at the mental health ball. This Cinderella metaphor has returned repeatedly, with Tarjan (1966) and King, State, Shah, Davanzo, and Dykens (1997) predicting that in the not-too-distant future, intellectual disabilities would soon gain in prominence within the mental health field.

Although one might be skeptical of a subfield whose emergence is repeatedly promised over a 91-year span, our sense is that, this time, the study of intellectual disabilities has arrived. Over the past two decades, the field has featured an increased number of studies (Hodapp, Fidler, & Depta, 2016), as well as new subdisciplines and journals focusing on policy (*Inclusion*; *Journal of Policy and Practice in Intellectual Disabilities*), autism (*Autism Research*; *Research in Autism Spectrum Disorders*), neurodevelopmental disorders (*Journal of Neurodevelopmental Disorders*), and health and mental health (*Disability and Health Journal*; *Journal of Mental Health Research in Intellectual Disabilities*). In addition, professionals in several disciplines now routinely study aspects of persons with intellectual disabilities. Developmental psychologists are intrigued by how children with some syndromes show wide discrepancies in their abilities in certain areas compared to others; geneticists, by how newly discovered genetic mechanisms are revealed in specific genetic syndromes; and mental health professionals, by how some mothers, fathers, and siblings cope well, whereas others have difficulties. Nurses, pediatricians, early interventionists, special educators, developmental neurologists, and neuropsychologists—all have their profession-specific reasons for being interested in intellectual disabilities.

For infant mental health professionals as well, intellectual disabilities present at least four interesting issues. First, we briefly describe the definition–classification, assessment, and diagnosis of infants and young children with disabilities. Second, we describe an "etiology-based" approach, using as examples the early development of young children with Down syndrome and Williams syndrome. Third, we address family functioning, including work examining functioning in families of children with

different genetic conditions. Finally, we review early intervention services and systems.

Definition

Listed in DSM-5 as the first of the neurodevelopmental disorders, intellectual disability "is a disorder with onset during the developmental period that includes both intellectual and adaptive functioning deficits in conceptual, social, and practical domains" (American Psychiatric Association, 2013, p. 33). The DSM-5 definition, which has remained substantially unchanged for over 50 years, thus focuses on the core features of (1) intellectual deficits, (2) adaptive functioning deficits, and (3) onset of problems during the childhood years (American Psychiatric Association, 2013).

Although the age criterion is relatively straightforward, the first two of these diagnostic criteria have been trickier to operationalize. "Intellectual deficits" generally refer to a child's performance on individually administered, recent, and well-standardized IQ tests. Given the error band of most IQ tests, "on tests with a standard deviation of 15 and a mean of 100, this involves a score of 65–75 (70 ± 5)" (American Psychiatric Association, 2013, p. 37). In general, though, intellectual disabilities occur when the person's IQ is below 70.

The second criterion relates to deficits in adaptive functioning. Most professionals view adaptive behavior as one's ability to perform everyday activities of daily living (e.g., eating, grooming, toileting), to communicate and socialize with others, and to understand social rules (Carter, Marakovitz, & Sparrow, 2006). While IQ has historically been emphasized, only those children with both IQ and adaptive deficits should be diagnosed with intellectual disabilities.

Classification

Traditionally, children have been classified as having mild (IQ 55–69), moderate (IQ 40–54), severe (IQ 25–39), and profound (IQ < 25) intellectual disabilities, with each successive level referring to children with progressively lower IQ scores. This "degree of impairment" approach to classification has been somewhat supplanted by a more functional approach. This nosology is based on the idea that some individuals may require only intermittent environmental supports, whereas others require limited, extensive, or pervasive supports (Schalock et al., 2010). In general, then, professionals differentiate children with intellectual disabilities on the basis of the severity of their impairment, measurable by their IQ level, functional and adaptive behavior, or their need for environmental supports.

In recent years, however, another approach to classification has become prominent. This approach classifies individuals based on the specific cause or etiology of their intellectual disabilities. An early version of this etiology-based approach can be seen in Zigler's (1967) "two-group approach" to intellectual disabilities, as it divided persons with intellectual disabilities into those with one or more clear organic causes ("organic" intellectual disabilities) or those showing no clear organic cause ("nonspecific" or "familial" intellectual disabilities). Given recent advances in molecular and clinical genetics, we are now able to diagnose many more children with a clear organic—usually genetic—cause. Many of the most exciting discoveries of the past two decades have related to children with genetic conditions of intellectual disabilities.

Assessment

Traditionally, assessment of intellectual disabilities has relied on standardized instruments of both intelligence and of adaptive behavior. Past the infant and toddler years, most clinicians rely on the Stanford–Binet or the Wechsler-based tests to assess intellectual abilities, and the global IQ score serves as the criterion against which the child is evaluated. Adaptive behavior is similarly examined with the Vineland Adaptive Behavior Scales or other adaptive scales, although clinician judgment is more often allowed in this area. In both cases, the "gold standard" relates to psychometric instruments that are well standardized and of recent vintage, and that provide overall standard scores.

Although reliance on overall IQ scores derived from standardized tests continues to predominate, the testing world has increasingly moved toward a more multifaceted view of intelligence (Sparrow & Davis, 2000). Most newly designed tests differentiate IQ into multiple domains, and such domains are thought to better reflect the true nature of intellectual function-

ing. Even in infancy, the Bayley Scales of Infant Development—Third Edition (BSID-III), divide intelligence into five separate domains. Similarly, most tests for toddlers and young children differentiate intelligence into multiple domains (Lichtenberger, 2005). Although this more differentiated view of intelligence has not always influenced diagnosis or intervention, it does relate to many of the major advances within the intellectual disabilities field.

Diagnostic Challenges

Even with recent advances in intellectual and adaptive testing, accurate diagnosis of intellectual disabilities remains challenging during the infancy or early childhood period. Parents do not always know what is "normal" for young children of different ages, and many infants simply do not come to the attention of early interventionist, infant mental health, or other professionals. Partly for this reason, the prevalence rates of children with intellectual disabilities rise from the preschool into the school years (Roeleveld, Zielhuis, & Gabreels, 1997; Westerinen, Kaski, Vita, Almqvist, & Iivanainen, 2014), the point at which children are identified though the educational system. Additionally, clinicians may be more apt to consider such diagnoses as specific language impairments or autism spectrum disorder, which may or may not ultimately be associated with intellectual disabilities.

Developmental screeners administered by pediatricians or nurses, and public health campaigns such as "know the signs" for autism, can identify children showing delays in motor, language, or social development, but the diagnosis of a full-blown intellectual disability is complicated by several factors. The primary, inherent difficulty in diagnosing intellectual disability during infancy is the instability of infant IQ scores from one time point to the next. Bayley developmental quotient (DQ) scores taken at 1 year of age generally show relatively little stability with scores on the Stanford–Binet or other childhood IQ tests when the same children are 3–5 years old; test–retest correlations are as low as 0 to .15 (Molfese & Acheson, 1997). This lack of predictive validity is believed to occur because "development occurs so rapidly and because test items may measure different constructs at different ages" (Lichtenberger, 2005, p. 198).

Instability of IQ scores, however, may not hold for infants and young children with more severe levels of impairment. Examining infants (average age 11 months) with developmental delays, Maisto and German (1986) noted that correlations between DQ and IQ scores until retesting up to 4 years later were moderately high, with most test–retest correlations ranging from .40 to .60. Other studies have found similar moderate correlations of early to later IQ scores, particularly among those young children who score at lower IQ levels (e.g., Jary, Kmita, & Whitelaw, 2011; Niccols & Latchman, 2002). Children with IQs below 50 may have even more stable IQ scores over time.

Other issues relate to the tests themselves. Although calls have repeatedly been made for improved IQ tests during infancy and early childhood, certain problems remain. For instance, historically, the Bayley Scales have provided DQ scores only down to DQ 50 (although the BSID-III does go down to DQ = 40), with lower functioning simply denoted as "DQ less than" this lowest value. Partly as a result, over the years, several attempts have been made to "extend the curve" to lower DQs, but the psychometrics of such downward extensions remain unknown.

Thus, the instability of IQ and the difficulties in assessing such rapid development in very young children serve to complicate the diagnosis of intellectual disabilities in infants and young children. These complexities seem particularly striking in young children with mild delays; these children often are from lower socioeconomic status (SES) or minority groups, and may or may not show concurrent deficits in adaptive behavior (Gresham, MacMillan, & Bocian, 1996). Increasingly, these more mildly affected children are diagnosed with learning disabilities, especially as they get older (MacMillan, Gresham, & Bocian, 1998). Much less diagnostic controversy occurs when infants show more severe impairments.

In the same way, many fewer controversies relate to children who show clear causes for their intellectual disabilities. Children with Down syndrome, Williams syndrome, fragile X syndrome, and nongenetic disorders, such as fetal alcohol syndrome, all show deficits that are easier to identify and whose developmental course is easier to track. Many of the field's most intriguing findings concern the early development of these children.

Etiology

From 750 to 1,000 genetic disorders have now been associated with intellectual disability (Ellison, Rosenfeld, & Schaffer, 2013), and individuals with genetic syndromes constitute a substantial percentage of those with intellectual disabilities (probably about 40%), with higher percentages at the lower IQ levels (Heikura et al., 2005). Most etiologies involve chromosomal abnormalities such as "trisomies" (i.e., having three copies of the same chromosome), "deletions" (i.e., part of a chromosome absent), "translocations" (i.e., part of a chromosome out of place), or "uniparental disomies" (two copies of the same chromosome from one parent and no copy from the other parent; Biringen, Fidler, Barrett, & Kubicek, 2005). Researchers are increasingly performing molecular- and mouse-model studies to examine the function of genes associated with a specific genetic condition; these studies shed light on biological mechanisms operating along the pathways from genes to brain to behavior. Neuroimaging studies have implicated particular brain regions in some disorders (Schaer & Eliez, 2007).

Such advances are reflected in increased medical scrutiny of infants suspected of having a genetic disorder. Infants with genetic diagnoses may show obvious or subtle physical, facial, or behavioral signs, or may have demonstrable impairments in cardiac, muscular, neurological, motor, or sensory systems. Guidelines have been developed that assist clinicians in knowing when to send infants for genetic testing (Curry et al., 1997), as have syndrome-specific recommendations for medical best practices in infants, children, and adults (e.g., Bull & the Committee on Genetics, 2011; Goldstone, Holland, Hauffa, Hokken-Koelega, & Tauber, 2008). Because genetic disorders often make infants more prone to specific health conditions, knowing about such conditions and medical best practices allow for ongoing vigilance and prompt care of the child's variable health needs (Intellectual and Developmental Disabilities Toolkit, 2017; Roizen, 2010). See Table 21.1 for an example of common medical conditions in young children with Down syndrome.

Knowing the genetic etiology is also important because infants with different genetic disorders are prone to show specific strengths and weaknesses in social, cognitive, linguistic, emotional, and motor domains; many also show particular maladaptive behaviors (Dykens, Hodapp, & Finucane, 2000; Hodapp & Dykens, 2012). In addition to profiles, children with a particular disorder may have periods when development is faster or slower. Such developmental information is increasingly being used in early intervention efforts (Fidler, Philofsky, & Hepburn, 2007; Hodapp & Fisher, 2017).

In addition, etiology-based information can help couples in their reproductive and family planning. The presence of certain genetic disorders (e.g., fragile X syndrome) indicates increased risk for future children in the family or, in some cases, for other relatives who may not know that they or their children are at risk. In contrast, other abnormalities are not likely to reoccur within the same family (e.g., most cases of Prader–Willi syndrome). In these cases, families have no greater risks of having a second

TABLE 21.1. Common Health Problems in Infants and Young Children with Down Syndrome

- Congenital heart defects (approximately 50%; perform echocardiogram)
- Celiac disease (3–10%)
- Congenital gastrointestinal defects (in 5–10% of infants)
- Hypothyroidism (50% or more; screen at 6 and 12 months, annually thereafter)
- Hearing problems (two-thirds of children ages 2 months–3 years had unilateral or bilateral hearing loss)
- Vision problems (46–100%, including cataracts from birth in 5%)
- Leukemia (cumulative risk by 5 years = 2.1%; perform complete blood count)
- Seizures (8% of children and adolescents, with 40% of seizures present before 1 year)
- Respiratory problems (one-half to two-thirds hospitalized for pneumonia, bronchitis, and bronchiolitis in first 2 years of life)

Note. Information about conditions from Roizen (2010), including evaluation, prevention, and surveillance guidelines from Roizen's Table 1 (pp. 24–25).

child with a specific disorder than do others in the general population.

Genetic Disorders

Although a comprehensive review of genetic disorders is beyond the scope of this chapter, we briefly describe two disorders: Down syndrome and Williams syndrome. Both are noteworthy in that they are fairly common, show distinct behavioral profiles, and feature a fair amount of developmental and behavioral research. In these ways, they demonstrate the promise of research on infants and young children with genetic etiologies.

Down Syndrome

Occurring in approximately 1 per 800–1,000 live births, Down syndrome is the most common chromosomal disorder involving intellectual disability. However, numbers differ based on maternal age, with the risk of having a child with trisomy 21 increasing from 1 in 1,000 at maternal age 30 to 9 in 1,000 at maternal age 40 (Hook, 1981). In the large majority of cases, Down syndrome is caused by having three copies of chromosome 21 (so-called "trisomy 21"). Children and adults with this disorder also often display several physical characteristics, including short stature, short broad hands, and distinctive facial features, such as a flat nasal bridge and a protruding tongue. Congenital heart defects (particularly of the atrioventricular canal) appear in approximately 50% of these infants. Significant hearing loss is present in approximately 90% of individuals with Down syndrome and may contribute to learning deficits and lower IQ (Mazzoni, Ackley, & Nash, 1994). Within infancy, half or more of these children have one or more hospitalizations following birth, often for pneumonia, bronchitis, and other respiratory problems (Boulet, Molinari, Grosse, Honein, & Correa-Villaseñor, 2008; So, Urbano, & Hodapp, 2007).

Over the past few decades, geneticists have identified a "critical region" of chromosome 21 that appears to be responsible for the intellectual disabilities and most of the facial features associated with Down syndrome (Korenberg, 1993). The Down syndrome critical region gene 1 (*DSCR1*) has also been implicated in cardiac defects and intellectual disabilities. An increased risk of leukemia in patients with Down syndrome has been linked to an interaction between a gene in the critical region (*FMS*-related tyrosine kinase 3, or *FLT3*) and a gene located on chromosome 3 (*GATA*-binding protein 1) (Look, 2002; Wechsler et al., 2002).

Although prenatal screening has become an established part of obstetric practice, the advent of noninvasive prenatal testing (NIPT) has dramatically altered the landscape of prenatal testing for Down syndrome and other aneuploidies (Murdoch et al., 2017). NIPT technologies analyze the circulating, cell-free fetal DNA present in a maternal blood sample. The sensitivity and specificity of NIPT for Down syndrome is 99%, making it an appealing alternative to less accurate or more invasive approaches. NIPT is expanding rapidly, even though the implications have yet to be developed for professional practices, counseling, and families.

Although in psychological research Down syndrome is often used as the "control" or "contrast" condition for children with Williams syndrome or autism, children with Down syndrome do have their own characteristic behavioral features. Behaviorally, most children with Down syndrome score in the moderate range of intelligence (IQ 40–54), although IQ scores vary widely from one child to another. Most studies show that these children display their highest IQ scores in the earlier years, with gradually decreasing IQs as they age (Hodapp, Evans, & Gray, 1999). Even during the earliest years, infants and young children with Down syndrome slow in their development as they get older (Dunst, 1990).

Young children with Down syndrome also show an etiology-related profile of strengths and weaknesses. Miller (1999) noted that, across the preschool period, most children with Down syndrome showed a profile in which abilities in receptive language were advanced over the child's expressive abilities (and over the child's overall levels of mental abilities). Such discrepancies became more pronounced—for increasing numbers of children—as the study examined children over the preschool period. This pattern of receptive over expressive language abilities may also relate to the high rates of articulation problems among children with Down syndrome, as well as specific problems in linguistic grammar (McDuffie & Abbeduto, 2009).

Conversely, as a group, children with Down syndrome are considered by others to have strengths in social skills, at least at the basic

level. Thus, toddlers with Down syndrome look to others (as opposed to objects) more often (Kasari, Mundy, Yirmiya, & Sigman, 1990), and while performing problem-solving tasks at later ages, these children tend to look to adults and engage in social behaviors (Kasari & Freeman, 2001; Pitcairn & Wishart, 1994). At the same time, however, children with Down syndrome do not perform well on higher-level social tasks. For example, these children perform poorly on tasks of emotion recognition (Kasari, Freeman, & Hughes, 2001), and the levels they reach on theory-of-mind tasks are no better than their overall mental abilities (Abbeduto et al., 2006). In short, while infants and young children with Down syndrome are oriented toward others, their "sociability" is confined mostly to the lowest levels of social skills (Cebula, Moore, & Wishart, 2010).

Recent work combines cognitive–linguistic weaknesses with infant–toddler sociability. By examining the early development of infant cognitive skills and infant behaviors during mother–child interactions, Fidler, Philofsky, Hepburn, and Rogers (2005) found that infants with Down syndrome show particular difficulties in means–ends (i.e., instrumental) thinking, or tasks that involve the idea that objects (e.g., stick, stool) can be used as a means for obtaining something desired. Such deficits seem to relate to these children's increased amounts of looking to others for solutions to difficult problems. Eventually, "the coupling of poor strategic thinking [i.e., poor means–ends thinking] and strengths in social relatedness is hypothesized to lead to the less persistent and overly social personality–motivational orientation observed in this population" (Fidler, 2006, p. 147). Although much remains to be discovered, early developments may hold the key to understanding many etiology-related behaviors in Down syndrome.

Williams Syndrome

Occurring in approximately 1 per 10,000 live births, Williams syndrome is caused by a microdeletion on chromosome 7 that contains approximately 25 genes. Children and adults with this disorder have a particular facial appearance, with a small "pug" nose (what used to be called an "elfin-like" appearance). Cardiac abnormalities (especially supravalvular aortic stenosis) are present in about 80% of children with Williams syndrome.

Behaviorally, most children with Williams syndrome score in the mild range of intellectual disabilities (IQ = 55–69) (Howlin, Davies, & Udwin, 1998), and these scores remain stable throughout adulthood (Fisher, Lense, & Dykens, 2016; Searcy et al., 2004). Most children with Williams syndrome show relative strengths in language; in fact, early researchers argued that children with Williams syndrome might have near-normal or "spared" levels of language. Although such spared language is now known to occur in only a few persons with Williams syndrome (Bishop, 1999), these children's levels in language and communication do appear higher than (but correlated with) their overall mental abilities (Mervis, 2012). Conversely, visual–spatial processing skills appear particularly weak, such that children with Williams syndrome have extreme difficulty in drawing pictures, distinguishing left from right, and performing other visual–spatial tasks (Dykens, Rosner, & Ly, 2000). Finally, in addition to friendly—even overly friendly—personalities, most children with Williams syndrome are extremely fearful and anxious (Dykens, 2003; Einfeld, Tonge, & Florio, 1997). From an early age, many children with Williams syndrome display a strong attraction to music. In addition to instinctively using music in a therapeutic manner to reduce anxiety and to increase positive affect (Dykens, 2003), the neuropsychological correlates of such reactions are increasingly being examined (Lense, Gordon, Key, & Dykens, 2014).

In addition to such behavioral profiles, three aspects of Williams syndrome are particularly noteworthy. First, researchers have begun to tie specific gene regions to particular physical and behavioral characteristics. The gene for elastin (*ELN*), for example, is known to be responsible for the cardiac defects and some craniofacial abnormalities. Deficits in visual–spatial construction, learning, and memory have been associated with lim domain kinase 1 gene (*LIMK1*; Frangiskakis et al., 1996), and general transcription factor genes (*GTF2I, GTF2IRDI*) have been associated with general intellectual disability and visual–spatial deficits in Williams syndrome (Morris et al., 2003).

Second, early diagnosis of Williams syndrome now seems possible based on both physical and medical characteristics. Until now, diagnosis during early childhood was difficult due to phenotypic variability and the subtle nature of the syndrome's physical characteristics.

Indeed, most infants with Williams syndrome first present with cardiovascular abnormalities or with prolonged colic (sometimes lasting a year). Huang, Sadler, O'Riordan, and Robin (2002) showed that although, on average, parents reported first being concerned about their child's health or behavior at a median age of 12 months, the diagnosis of Williams syndrome was not made until age 3.66 years. But by including a clinical geneticist in the evaluation process, the mean delay in diagnosis was reduced almost in half, and the number of diagnostic tests performed dropped from 8.2 to 5.2.

A third advance relates to early development within Williams syndrome and some glimmerings of the later-emerging phenotype. In addition to work documenting infants' delays in pointing, showing, and other communicative gestures that typically precede early verbal language development (Mervis & Becerra, 2007), studies also document keen interests in faces and aberrant facial gaze (often peering at another person's face from close range) in toddlers with Williams syndrome (Laing et al., 2002). There may also be a connection between adaptive and maladaptive behaviors during the preschool years, such that levels of adaptive communication and socialization are inversely related to problem behaviors in social relating (Hahn, Fidler, & Hepburn, 2014). Future years promise even greater understanding of early development for children with Williams syndrome and other genetic intellectual disability syndromes.

Family Adaptation

The field of infant mental health has long been concerned with infants in the context of caregiving relationships, recognizing that infant and context reciprocally transact over time. Before describing some of the field's recent findings regarding families' adaptations, we provide a brief sense of the past, present, and emerging trends in this area.

Changing Perspectives

Research on families of children with disabilities features a clear change over time (Hodapp & Ly, 2005). Prior to the mid-1980s, the dominant perspective could be characterized as pathological or negative. Beginning with the seminal work of Solnit and Stark (1961), mothers were considered to mourn, as in a death, the birth of the newborn with disabilities. Mothers were thought to traverse a set of stages from shock, denial, and disbelief to anger or depression, to emotional reorganization. Although various theorists modified this stage-like process and stages did not always work as predicted (Blacher, 1984), the model was thought to characterize maternal reactions. Most early studies focused on mothers, although a few examined maladaptive behaviors–emotional problems of fathers (Friedrich & Friedrich, 1981) and siblings (Lobato, 1983). Families, overall, were also conceptualized as being at risk, with various studies focusing on economic immobility, marital conflict and divorce, and the inability of families to "grow up" with their children (particularly those with severe–profound intellectual disabilities; Farber, 1960).

Even though this exclusive focus on negative family outcomes predominated, most researchers appreciated that while some families experienced difficulties, others did not. But how did such "nonpathological" mothers, fathers, siblings, and families overall fit within what was mostly a negative and pathological view?

In 1983, Crnic, Friedrich, and Greenberg (1983) proposed a different way to conceptualize these families. According to these theorists, the child with disabilities was most appropriately considered as an added stressor in the family. Like the family's moving from one town to another, or the illnesses, promotions, or other changes that individual family members undergo, so too could the birth, diagnosis, and raising of the child with disabilities be considered as an added stressor. The experience of dealing with added stress, in turn, could be either detrimental or helpful to the family.

This changing perspective led to changes in both theory and practice. Theoretically, the stress-and-coping perspective led to interest in models that considered the correlates of family success. One such model was the Double ABCX, a revision of a family model originally employed by McCubbin and Patterson (1983). According to this model, the "crisis" (or "X" term) of raising a child with disabilities was dependent on characteristics of the child ("A"), the family's internal and external resources ("B"), and the family's perceptions of the child and the child's role in the family ("C"). The "double" conveyed the notion that child characteristics, family resources, and family perceptions of the child can all change over time. Although some-

what nonspecific, this type of model has been used successfully in understanding coping in families of children with disabilities (Minnes, 1988).

Practically, the stress-and-coping perspective and the Double ABCX model have changed the focus of interventions. First, not all parents and families need intervention. Many—even most—are coping well. In addition, by considering child and family factors that may lead to better and worse coping, interventionists can both screen and intervene more effectively. Emphasis also changes from considering families of children with versus without disabilities (between-group focus) to one that considers both between-group and within-group issues. By identifying which variables affect family functioning, within-group examinations can help identify which families are most likely to cope well, which are not, and why. Stress-and-coping theories provide a more nuanced, balanced account of these families.

Although an improvement on more negative conceptualizations of families, over the past decade, such stress-and-coping perspectives also have received criticism. Some of the following criticisms are not, by themselves, incompatible with stress-and-coping models, but they shift the focus more from deficits to benefits, and all will likely be receiving increased attention in the years to come.

New Directions

Positive Effects and Fewer "Clinical" Interventions

Many researchers feel that stress-and-coping models unduly focus on either negative effects or, at best, familial coping or "getting by." In many cases, however, families feel that they have benefited from raising a child with disabilities. Such benefits include appreciating life more, understanding life's deeper meanings, being more empathic and understanding of differences, feeling more committed to social justice, and helping those less fortunate than ourselves (Dykens, 2006). Although such benefits have only begun to be examined in families of children with disabilities (Taunt & Hastings, 2002), studies of such positive effects balance the existing body of negative (or at least neutral) studies.

Related to the issue of positive effects is the possibility that various "nonclinical" interventions can be helpful for these parents. One helpful intervention involves mindfulness-based stress reduction (MBSR). Long used with individuals with a host of conditions and undergoing various life traumas, MBSR has recently been shown effective for helping parents of children with disabilities (Dykens, Fisher, Taylor, Warren, & Miodrag, 2014). MBSR, positive parenting, and other supports and aids—as well as more traditional clinical interventions, when necessary—have helped many parents of children with disabilities (Dykens, 2015).

Increased Attention to Other Family Members

Although stress-and-coping models have been less "mother-centric" than earlier studies, to this day few studies examine other family members or families overall. We need sustained, long-term studies of fathers (MacDonald & Hastings, 2009); the family's economic mobility (Farber, 1960); and the amount, timing, and correlates of parental divorce (Risdal & Singer, 2004). And although there are many methodological challenges in studying the siblings of children with disabilities (Hodapp, Glidden, & Kaiser, 2005), the nature of sibling problems—and rewards—are beginning to be examined (Shivers & Dykens, 2017).

Focus on Etiology and Etiology-Related Family Outcomes

The cause or etiology of intellectual disabilities in the child may relate to family functioning. As compared to same-age children with other intellectual disabilities, for example, families of children with Down syndrome may cope better (Hodapp, Ly, Fidler, & Ricci, 2001; Seltzer & Ryff, 1994). Yet why such a "Down syndrome advantage" exists remains unclear. Parents may react positively to children who are more often socially oriented and who generally have lesser (though not absent) degrees of severe maladaptive behavior (Dykens, 2007). At the same time, however, parents of children with Down syndrome continue to be older (by about 5 years) than parents of typical children or children with other disorders; whereas this state of affairs may be more problematic when offspring with Down syndrome are themselves middle-aged (Hodapp, Burke, Finley, & Urbano, 2016), during childhood it may lead to a higher likelihood that parents have more education, are married, and provide more resources for their children (Hodapp et al., 2012). Beyond such parental–

familial issues, Down syndrome features more support groups and may also be a disorder that is more accepted by the public.

Increased Focus on Real-World Outcomes

Most recent family studies have focused on maternal stress. Although a few studies have examined telomere shortening (Epel et al., 2004) and sickness–hospitalization among parents of young children with disabilities (Miodrag, Burke, Tanner-Smith, & Hodapp, 2015), few examine other types of real-world outcomes. For example, few studies examine how often or when mothers or fathers get sick or are hospitalized, or how or when they decide to have more children, accept a promotion, go back for further schooling, change their life's work, or move to another house, town, or state. We know little about the brothers/sisters of children with disabilities, and whether being a sibling to a brother/sister with intellectual disabilities might influence such siblings in their academic, social, marital, or occupational choices and successes. Having so far focused on stress, the field now needs to focus on real-world outcomes for parents and siblings of a child with disabilities.

Summary of Family Findings

Overall, families of children with disabilities show only slight negative effects. In contrast to popular thinking, meta-analyses of several decades of studies show that, as a group, mothers suffer only slightly to moderately more depression than do mothers of same-age nondisabled children (Singer, 2006). Similarly, couples of children with disabilities show only small increases in their divorce rates, with families of children with disabilities showing divorce rates that are only 6% higher compared to control families (Risdal & Singer, 2004). Similarly for siblings, Rossiter and Sharpe (2001) found slight negative effects overall, with the largest negative effects arising when siblings of children with (vs. without) disabilities were examined using direct observation, and when outcomes related to anxiety and depression. Siblings of children with disabilities may also show slightly better social functioning.

Beyond these more general findings, correlates of coping are found both in mothers and in children with disabilities. Among mothers, the main correlate concerns the mother's coping style. In contrast to "palliative" coping, in which the mother either denies or perseverates on her negative emotions, mothers who engage in active, problem-solving coping do better (Essex, Seltzer, & Krauss, 1999). From the child's perspective, those children who show more behavioral problems, particularly acting-out behaviors, appear to be more difficult for parents to handle (Glidden & Schoolcraft, 2007). Compared to parents of same-age children with other disabilities, parents of children with autism may show more depression (Hayes & Watson, 2012; Singer, 2006).

Parents also experience more depression and higher stress levels during the early years, after the birth (or early diagnosis) of the child with disabilities. In Glidden and Schoolcraft's (2003) studies of adoptive versus birth parents of children with disabilities, the birth mothers experienced high levels of depression during the child's earliest years, even as the two groups were indistinguishable when children became older. The adverse effects of having a child with disabilities seem most pronounced during the infancy and early childhood period.

In addition, compared to mothers of children with other types of intellectual disability, mothers experience less parental stress when their children have Down syndrome (Hodapp, 2007). In one study examining administrative records of an entire state over a multiyear period, parents of children with Down syndrome divorced at slightly lower rates compared to parents of (nondisabled) children from the entire state population or to parents of newborns with congenital anomalies other than Down syndrome. In contrast, Hartley and colleagues (2010) noted that divorce rates were higher for parents of children with autism, which may be in line with the generally increased stress levels experienced by parents of these children (Hayes & Watson, 2012). Especially among the families of children with Down syndrome, couples were more prone to divorce if they married at younger ages, were less educated, and were both rural and less educated (Urbano & Hodapp, 2007).

Taken together, family studies have changed greatly over the past several decades. In addition to changing theoretical perspectives, recent studies have produced more precise findings that might lead to better screening and intervention. Granted, parents and families still experience periods of sadness and increased stress (possibly more often during the early years), but most families cope well, negative effects are (on

average) small, and many families find many positives in raising children with disabilities. Still, our knowledge of such families remains incomplete, even as we acknowledge the importance of families to the well-being and development of young children with disabilities.

Service Delivery

Within the U.S. system of special education services, services to infants and young children are a later-arriving offshoot. After a series of court decisions and legislative achievements during the 1960s and early 1970s, Public Law 94–142, the Education for All Handicapped Children Act, was passed in 1975. It codified the right of all U.S. children to a free, appropriate, public education within the least restrictive educational setting. No longer could any school-age child, whatever his or her disability, be denied access to public education. Known today as the Individuals with Disabilities Education Act (IDEA), this law was reenacted (with slight modifications) in 1997 and 2004 (Hallahan & Kaufman, 2006).

Public Law 94–142 and IDEA mandated services for school-age children, but not for children below age 3 years. Concern about such younger children led to the passage of Public Law 99–457 in 1986, including mandatory special education for children ages 3–5 years and a voluntary grant program for infants–toddlers below age 3 years. Although similar to IDEA, the early intervention services for infants and toddlers differ in ways other than simply serving younger children (Krauss & Hauser-Cram, 1992).

A first major difference involves who runs the services. Although school-age children's services are under the direction of the child's local school district, early intervention services are administered through the states. In each state, a state-designated "lead agency" directs early intervention services. This agency might be the state's department of education, health, or intellectual and developmental disabilities. All early intervention services are then coordinated by that lead agency.

A second difference relates to which children are eligible for services and the areas of intervention provided to each child. For school-age children, eligibility for special education services occurs when children have established disorders or diagnoses (as determined by appropriate tests and diagnostic procedures). In early intervention, infants–toddlers are eligible for services if they have development delays, or if they have conditions that involve established risks (e.g., Down syndrome, fragile X syndrome), biological risks (prematurity, low birthweight), or environmental risks (extreme poverty, homelessness, suffering from abuse or neglect). Such differences in eligibility criteria and in which children are served constitute a major change from later, school-based services (Buysse, Bernier, & McWilliam, 2002).

A third and striking difference concerns the family focus of early intervention services. With the exception of parental agreement concerning the child's individualized education plan (IEP), parents and families receive little attention in special education law and services. But reflecting the long-held view that infants and their families are inextricably linked, early intervention services strive to be family-centered. For example, early intervention services are based on the individualized family service plan (IFSP). This document, in addition to its descriptions of the young child's levels and needs, also includes a statement of the resources, priorities, and concerns of the child's family, along with statements about how early intervention services will meet the family's needs. Although debate continues concerning the degree to which early intervention services truly meet child and family needs (Raspa, Hebbeler, Bailey, & Scarborough, 2010), IFSPs and the field as a whole do provide clear plans for the provision of services.

Going beyond the IFSP document per se, early interventionists attempt to include parents and other family members in conceptualizing and intervening with young children with disabilities (Bruder, 2000). Trivette, Dunst, Boyd, and Hamby (1996) have even developed a typology of program types, ranging from "professionally centered" on one end to "family-focused" on the other. Differences relate to the degree to which professionals help the parents achieve their own goals and become full participants in the early intervention process. Still, the goal of fully including parents in early intervention is not always met. Despite what program practitioners say they do, parents generally report that most early intervention programs are not fully "family-centered" (Dunst, 2002). Although programs for infants (vs. for those preschoolers) more closely approximate family-centeredness (Dunst, 2002), the goal that parents should be

full participants in early intervention continues to be only sporadically realized.

Along with the problem of parent- and family-centered services, the early intervention field is also beginning to acknowledge other gaps in service delivery. In an article aptly subtitled "Powerful Vision and Pesky Details," Hebbeler, Spiker, and Kahn (2012) decry the many issues with which the field increasingly grapples. These details include questions of access, quality, and costs–funding. In the early intervention context, access involves problems in identifying and serving all eligible children, whereas quality relates to identifying and training qualified personnel, developing national standards for such personnel, providing high-quality services in inclusive settings, and allowing families to navigate more effectively the transition from early intervention to school-based services when the child turns 3 years of age. Another major issue concerns costs, especially the ways in which federal allocations for early intervention services have declined over the decades (Lazara, Danaher, & Goode, 2011). Each of these issues becomes exacerbated when (1) states are low in resources or face budget shortages (Cole, Oser, & Walsh, 2011); (2) children are more mildly impaired (in some states, they then are no longer eligible for early intervention services; Cole, Oser, & Walsh, 2011); and (3) parents–families speak Spanish or are poor (Williams, Perrigo, Banda, Matic, & Goldfarb, 2013).

Seen within this mixed picture of early intervention services and programs, most mental health professionals have nevertheless come to appreciate the importance of focusing on screening and intervention at the earliest age possible. As a result, early screening, diagnosis, and intervention have become routine for infants and young children who either have, or are at risk of having, intellectual disabilities and/or other psychiatric conditions. In addition, several initiatives have successfully trained community pediatricians to identify and refer for more specific testing those infants–toddlers who are suspected of having autism spectrum disorder (Swanson et al., 2014). More generally, whether parents and their young children arrive at a child guidance clinic, a pediatric or child psychiatry department in a large hospital, or a community mental health clinic, professionals agree that "the earlier the better" is best in terms of diagnosis and intervention. These early-diagnosed children and their families can then benefit from the entire gamut of early intervention services.

Concluding Thoughts

In considering the status of intellectual disabilities within the field of infant mental health, we see a field that is itself in its infancy. Many issues continue to be difficult to solve, for example, the basic issue of how to accurately test and diagnose infants or toddlers with intellectual disabilities. On other questions, more progress seems evident. Even compared to 30 years ago, we now know much more about many genetic causes of intellectual disabilities, including how infants with several of these disorders evolve in their behavioral profiles and how genetic anomalies tie together with brain and behavior. Our knowledge has similarly advanced concerning how families cope with young children with intellectual disabilities. In future years, we can reasonably hope that our knowledge concerning such families will better inform early intervention services, thereby leading to more optimal outcomes for infants with disabilities and their families.

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