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Review

Intellectual disability and its relationship to autism spectrum disorders

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ABSTRACT

Intellectual disability (ID) and autism spectrum disorders (ASDs) covary at very high rates. Similarly, greater severity of one of these two disorders appears to have effects on the other disorder on a host of factors. A good deal of research has appeared on the topic with respect to nosology, prevalence, adaptive functioning, challenging behaviors, and comorbid psychopathology. The purpose of this paper was to provide a critical review and status report on the research published on these topics. Current status and future directions for better understanding these two covarying disorders was reviewed along with a discussion of relevant strengths and weaknesses of the current body of research.

Contents

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Intellectual disabilities (IDs) are characterized by social, cognitive, and adaptive skill deficits (Downs, Downs, & Rau, 2008; Lifshitz, Merrick, & Morad, 2008; Matson, Dixon, Matson, & Logan, 2005; Matson, Smalls, Hampff, Smiroldo, & Anderson, 1998; Matson, Smiroldo, & Bamburg, 1998; Myrbakk & von Tetzchner, 2008; Thirion-Marissiaux & Nader-Gosbois, 2008; Yalon-Chamovitz & Weiss, 2008; Zayac & Johnston, 2008). Additionally, ID is often accompanied by stereotypies and challenging behaviors (Lee, Harrington, Chang, & Conners, 2008; Matson et al., 1997a). Comorbid psychopathologies of various types such as anxiety, depression, and schizophrenia may also be present (Cherry, Matson, & Paclawskyj, 1997; Holden & Gitlesen, 2008; Matson & Bamburg, 1998; Matson & Smiroldo, 1997; Matson, Smiroldo, Hamilton, & Baglio, 1997; McGillivray, McCabe, & Kershaw, 2008). All of these problems can be problematic for client and staff, and in some instances can be very debilitating. The disorders with the greatest overlap with ID are those in the autism spectrum (ASD; Matson et al., 1996; Wilkins & Matson, 2009). The purpose of this review will be to assess how these two conditions interact, and how they affect many of the disorders and behaviors noted above.

Hurley and Levitas (2007) speculate that much of the recent advance in ASD has been with persons who are intellectually normal. They further posit that as a result, many persons with ID may be overlooked with respect to co-occurring ASD. This situation is the case despite the knowledge that has existed for several decades that considerable overlap in ID and ASD exist (Bartak & Rutter, 1976). Persons with ID and ASD have different needs from individuals with ID or ASD alone (Carminati, Gerber, Baud, & Baud, 2007; Gilchrist et al., 2001). Thus, making this distinction is much more than an academic exercise. Furthermore, one of the outcome measures typically described in early intervention programs is the improvement of IQ (Matson & Smith, 2008; Sheinkopf & Siegel, 1998). Additionally, lower IQ has been an indicator of poorer prognosis for these early intensive plans (Ben Itzchack, Lahat, Burgin, & Zachor, 2008).

Three distinct groups are therefore present—persons with ID, persons with ASD, and persons with both ASD and ID (Noterdaeme & Euders, 2009). The task of the clinician is to distinguish between these groups. Scales have been developed that are specific to this task (de Bildt, Systema, Kraijer, & Minderaa, 2004; Kraijer & de Bildt, 2005; Matson, Wilkins, Boisjoli, & Smith, 2008). However, they are of recent origin and deal only with adults. At present, making clear distinction between symptoms in these groups is difficult. More scale development is needed and should be influenced by nosology studies as well. This latter topic will be addressed in some detail.

1. Nosology

Recently, researchers have attempted to establish common genetic or neurodevelopmental pathways for ID and ASD. The great deal of overlap between the two disorders suggests possible genetic similarities. Terminal deletion of the long arm of chromosome 2 for example, has been suggested (Galasso et al., 2008). Similarly, Laumonnier et al. (2006) suggest a functional deficit of the BKca channel. Furthermore, a 1.5 mb duplication on chromosome 16p 13.1 has been suggested as a potential contributor to autism (Ullman et al., 2007). Conversely, in a single case report, Depienne et al. (2007) found that a deletion at the 7q 11.23 in one of several genes may be related to delayed language and social interactions. However, at present, little data are available to draw any firm conclusions relative to these genetic data (Levine, Morrow, Berdichevsky, & Martin, 2007).

Many persons demonstrate that individuals with autism have patterns of intellectual functioning that are distinct to this group (Bartak, Rutter, & Cox, 1975; Lincoln, Courchesne, Kilman, Elmasian, & Allen, 1988; Matson, Wilkins, Smith & Ancona, 2008). Kaufman, Kaufman, and Lincoln (2000) summarize much of this data, showing strengths in the visual-motor area that are most pronounced in information, vocabulary and coding.

Various nosology factors show interesting patterns between ID and ASD as well. For example, the more severe the person's ID, the greater the likelihood of ASD (Vig & Jedrysek, 1999). Whereas social competency deficits were marked in children with ASD, children with mild ID or typical development did not differ (Ingram, Mayes, Troxell, & Calhoun, 2007). Persons with autism who have ID also differ from persons with autism not only on core symptoms but also on co-occurring comorbid problems as well. For example, persons with lower IQ evinced significantly higher rates

of stereotypies and self-injury (Bartak & Rutter, 1976). Munson et al. (2008), who studied 456 children with autism, concluded that they had multiple IQ-based subgroups differing in cognitive strengths and weaknesses, and severity of autistic symptoms. Finally, Deb and Prasad (1994) found that impaired verbal and non-verbal communication and the occurrence of repetitive and restrictive activities were more common in persons with autism and ID versus autism alone.

When comparing persons with autism, with or without ID, to an ID group, the differences are much clearer. Jackson et al. (2003) for example found that in social interactions children with autism produced fewer positive responses and more no responses to verbal solicitations. Mandelbaum et al. (2006) compared 242 children 7–9 years of age who fell into one of four groups; high IQ and autism, developmental language disorder resulted in similar patterns of performance while low IQ and autism or low IQ were similar but differed from the two former groups. The two former groups had better sensory motor skills, oromotor skills, praxis and ability/willingness to perform tasks.

2. Prevalence

A substantial number of epidemiological studies have been done on the prevalence of ASD. However, due to changing criteria, better and better funded screening methods and other factors, rates have changed considerably (Newschaffer, Falb, & Gurney, 2005). One explanation is diagnostic substitution. Researchers have found that as the rate of ASD diagnoses go up, the rates of ID and learning disability diagnoses go down (Shattuck, 2006). Furthermore, comorbidity of ID and ASD can account for some of this increase. Children initially identified with ID only may now be viewed as evincing ID and ASD. For example, Sikora, Pettit-Kekel, Penfield, Merkens, and Steiner (2006) note that in one genetic form of ID, Smith-Lemil-Opitz Syndrome, all the cases they examined displayed either autism or pervasive developmental disorder not otherwise specified (PDD-NOS). Others have hypothesized that a greater portion of children who are intellectually more able are being diagnosed with ASD (Keen & Ward, 2004). Keen and Ward (2004) also speculate that this may at least in part account for the high overlap of ASD with attention deficit/hyperactivity disorder (ADHD).

Fombonne (2003) provides an excellent overview of many of these studies. What we do know from this review and from the studies themselves is that rates of Asperger syndrome, autism, and PDD-NOS are high relative to Rett's syndrome and childhood disintegrative disorder, which are very rare. Current estimates are that ASD occur in 1 of 150 children. Rates of individuals with ASD and ID are obviously lower and are about 50–70% of all ASD cases. For persons with ID, ADHD was the most common co-occurring condition, with ASD being the second most common co-occurring disorder (Stomme & Diseth, 2000). This finding was also reported for persons with ASD and "borderline" ID (IQ of 71–84) (Artigas-Pallares, Rigau-Ratera, & Garcia-Nonell, 2007).

LaMalfa, Lassi, Bertelli, Salvini, and Placidi (2004) conclude that 40% of persons with ID have an ASD, while 70% of persons with ASD have ID. Bryson, Bradley, Thompson, and Wainwright (2008) found that 28% of individuals with ID also evinced autism. de Bildt et al. (2004) reported much lower rates of overlap in ID and ASD. Using *DSM-IV-TR* criteria for Pervasive Developmental Disorder, they reported a 16.7% prevalence rate of comorbidity. Similarly, Wing and Gould (1979) reported that 11% of the severely ID children they studied were autistic. In a later study with adults who evinced ID, they reported a 4% rate of autism (Shah, Holmes, & Wing, 1982).

The prevalence data shows widely discrepant numbers in overlap between ID and ASD. Older studies generally show lower percentages of overlap. Expanded criteria for ASD may account, at least in part, for these findings, along with different types of samples (e.g., children versus adults, in patients versus outpatients, and different assessment methods). Also, the notion of comorbid disorders is taking on greater emphasis and may account for increasing prevalence rates between the two conditions. It should also be emphasized that most studies only refer to autism (versus other ASD in co-occurrence with ID). The addition of PDD-NOS, and the low incidence of Rett syndrome and childhood disintegrative disorder, would increase the percent of individuals with overlapping ID and ASD. These studies have yet to be conducted.

3. Adaptive behavior

The combination of ID and ASD presents many challenges and deficits across a range of behaviors and skills that are not seen in ID or ASD alone (Boucher, Bigham, Mayes, & Muskett, 2008). Most of the research to date on adaptive behavior has used the Vineland Adaptive Behavior Scale as the primary outcome variable. The focus thus has primarily been on looking at personal behaviors that allow the individual to be more self-sufficient. Schatz and Hamdan-Allen (1995) found that with increasing IQ, children with autism evinced less improvement in social and daily living skills. In a replication and extension of this study, Bolte and Poustka (2002) evaluated the association of adaptive behavior (using the Vineland Adaptive Behavior Scale) in autism or ID and PDD-NOS. Lower IQ was a primary mediator for ASD. Additionally, in persons with severe and profound ID, individuals with autism as a comorbid disorder had much greater deficits in social and adaptive behavior than persons whose comorbid disorder with ID was psychosis (Matson, Mayville, Lott, Bielecki, & Logan, 2003). Finally, Fodstad and Matson (2008) found that adults with severe and profound ID with ASD had more behaviorally based feeding problems, particularly food selectivity and refusal, compared to persons with ID alone.

The amount of research on adaptive behavior and the comorbid interactions of ID and ASD have received limited attention. However, these factors do appear to affect a host of adaptive behaviors and have important implications for treatment planning and independent living. More systematic research across levels of ID, and ASD across the lifespan is needed. Additionally, given the focus in the developmental disabilities field, it is interesting that more focus on identifying areas for skill building programs on social and adaptive behaviors have not been conducted (Matson, Carlisle, & Bamburg, 1998; Matson, Dempsey, & LoVullo, 2009).

4. Challenging behaviors

The study of challenging behaviors in ID and ASD is a topic that has recently seen a good deal of attention (Dawson, Matson, & Cherry, 1998; Hill & Furniss, 2006; Matson, Kiely, & Bamburg, 1997). Researchers have found for example that as IQ goes down, the severity of ASD and challenging behaviors goes up, making ID and ASD major risk factors (O'Brien & Pearson, 2004). Murphy, Healy, and Leader (2009) look at other potential risk factors for challenging behaviors. Age and gender were not related to these behavior problems despite the fact that 82% of their sample of 744 persons studied evinced challenging behaviors. They also, like Murphy et al. (2009), found that severe IQ and ASD were related to higher rates of challenging behaviors, but not across the board. Self-injury in particular was higher but aggression and stereotypies were not related to level of IQ in ASD. High rates of stereotypies tended to be related to severity of autism (Goldmon et al., 2009). For children with ID and autism, functional assessment for challenging behaviors showed that these behaviors were maintained by escape or the retention of tangible items (Reese, Richman, Belmont, & Morse, 2005).

Children with ID and ASD tend not to "grow out" of these challenging behaviors. Rather, they persist over time with persons with the highest rates of challenging behaviors still having the highest rates later in life. Murphy et al. (2005) observed this phenomenon in 141 persons with severe ID and autism at a 12-year follow-up.

Patterns of challenging behaviors should be a high priority for those who serve individuals with ID and ASD. These problem behaviors occur at very high rates, thus much is yet to be learned. These initial data are promising and should be helpful for diagnostic and treatment planning purposes.

5. Comorbid psychopathology

Evidence of comorbid mental health disorders in persons with ID and ASD is growing (Gadow, Devincent, Pomeroy, & Azizian, 2005; McCarthy, 2007). Hill and Furniss (2006) for example, assessed 82 persons with severe ID with and without autism. In comparing persons with severe autistic symptoms and ID to moderate autism to no autistic symptoms, the following pattern emerged. The most common comorbid disorders, in order of frequency were anxiety, mood disorder, mania, schizophrenia, and problem behaviors of impulse control and stereotypies. These symptoms were evident in severe autism

with the fewest symptoms in the ID only group. Symptoms of psychopathology in autistic individuals persist over time (Sabaratnam, Murthy, Wijeratne, Buckingham, & Payne, 2003). Often developing in early childhood, comorbid psychopathology is significantly higher in autism than for children with ID alone (Brereton, Tonge, & Einfeld, 2006). As previously mentioned, a range of comorbid psychopathology is present in persons with ID and ASD. Bakken, Friis, Lovoll, Smeby, and Martinsen (2007) studied 43 adults, all of whom had ID and autism and a formerly diagnosed mental health disorder. They found that 8 of those persons' mental health condition were psychosis.

Niklasson, Rasmussen, Oskarsdottir, and Gilberg (2009) studied 100 consecutive persons with 22q 11 deletion syndrome. ADHD was diagnosed in 51 of the cases. ASD was diagnosed in 23 cases (only 5 were autistic). In 9 of these cases with ASD, ADHD was also diagnosed. Bradley and Issacs (2006) also observed higher rates of inattention, hyperactivity, and impulse behaviors for teens with autism and ID compared to persons with ID alone. Finally, Bradley, Summers, Wood, and Bryson (2004) found that persons with ID and autism were more likely to evince psychopathology than persons with ID only. The ID plus autism sample was vulnerable to anxiety, mood, sleep problems, organic syndrome, and stereotypies and tics.

Bradley and Bolton (2006) looked at teenagers who had ID, with and without autism. They found that significantly more people with autism had a lifelong comorbid emotional disorder. Common comorbid disorders they observed were depression and bipolar disorder. Tsakanikos, Sturmey, Costello, Holt, and Bouras (2007) in a sample of 137 adults with ID and ASD, found comorbid psychopathology in 42% of the cases. The most frequently diagnosed disorder was schizophrenia, followed by depression, adjustment reaction, and anxiety. Having said that, researchers have been delayed in part due to a lack of adequate diagnostic methods (Hagopian & Jennett, 2008; MacNeil, Lopes, & Minnes, 2009). Furthermore, MacNeil et al. (2009) reviewed 13 studies where anxiety disorders were studied in ASD. However, participants invariably had Asperger's Syndrome or high functioning autism, which may suggest that the type or frequency of comorbid psychopathology in ASD may differ based on whether ID is present and its severity.

Comorbid psychopathology appears to be a set of conditions to which persons with ID and ASD are particularly vulnerable. Researchers are beginning to pay closer attention to these problems and the number of studies is beginning to increase. One reason for the lack of good prevalence and nosological data has been the lack of scales designed specifically to evaluate comorbid psychopathology in persons with ID and autism. This issue is now beginning to be addressed (Helverschou, Bakken, & Martinsen, 2009; Matson & Boisjoli, 2008). These developments suggest that this topic may pick up further momentum from researchers in the near future.

6. Conclusions

The study of ASD in persons with ID is a relatively new phenomenon. Until recently, the disorder was considered to be a condition of childhood. Furthermore, the possibility of ID was not factored into the diagnostic profile (Bartak & Rutter, 1976). In the last decade, the additive effect of related disorders and behaviors to the core ASD and ID symptoms has begun to receive increasing attention. This situation is likely due to the success of treatments across the lifespan. Furthermore, it is recognized that without effective interventions, these co-occurring conditions tend to persist.

ID is at the same time perhaps the most common co-occurring disorder with ASD, and a strong predictor of poor prognosis. This combination of factors suggests the need for even greater efforts at better understanding these two phenomena and their relationship to each other. At present, some useful information on nosology, prevalence, adaptive behavior, challenging behaviors, and comorbid psychopathology has begun to emerge. Assessment instruments specific to ID and ASD are starting to appear and should further aid in our understanding of the topics just mentioned. Study of these phenomena across the lifespan is needed as well as a host of other topics such as cognition, learning styles, communication, social skills, perspective taking, and a variety of other factors that affect quality of life and independent living. What is evident is that this ID and ASD group are distinctly different from persons with ID or with normal IQ and ASD. The available research on the topic is small at present, but promising and illuminating. Greater research efforts in the future are welcome and warranted.

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