Original Article

Using adaptive behavior scores to convey level of functioning in children with autism spectrum disorder: Evidence from the Study to Explore Early Development

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Abstract

We examined relationships between measures of adaptive behavior, cognitive ability, and autism symptom severity in 1458 preschool-aged children with autism from the Study to Explore Early Development. While publications commonly describe autistic children as "low-" or "high-functioning" based on cognitive ability, relying solely on cognitive scores may obscure meaningful variation in functioning. We found significant heterogeneity in adaptive behavior scores of children with cognitive scores both above and below the threshold of two or more standard deviations below the population mean specified in the diagnostic criteria for intellectual disability (ID). Although cognitive and adaptive behavior scores were strongly associated in our sample, considerable variation in overall adaptive behavior and more than half in socialization and motor skills was unaccounted for by cognitive ability, autism symptom severity, and other covariates. Among children who could be designated "low-functioning" based on cognitive scores, 39.7% had composite adaptive behavior scores indicating no significant delays, while among those who might be designated "high-functioning," 9.0% had significant delays in overall adaptive behavior and 22.2% in socialization. These results suggest adaptive behavior scores capture variations in the autism phenotype not accounted for by other measures we considered.

Lay Abstract

Autistic people are often described as "low-" or "high-functioning" based on their scores on cognitive tests. These terms are common in publications and in everyday communication. However, recent research and feedback from the autistic community suggests that relying on cognitive ability alone to describe functioning may miss meaningful differences in the abilities of autistic children and adults and in the kinds of support they may need. Additional methods are needed to describe "functioning" in autistic children. We examined whether scores from a test measuring adaptive behaviors would provide information on the functional abilities of children with autism that is different from cognitive ability and autism symptom severity. Adaptive behaviors include age-appropriate skills that allow people to function in their everyday lives and social interactions. We found that a large amount of the variation in adaptive behavior scores was not explained by cognitive development, autism symptom severity, and behavioral and emotional problems. In addition, there was a wide range of adaptive ability levels in children with autism in our study, including in those with low, average, or high cognitive scores. Our results suggest that adaptive behavior scores could provide useful information about the strengths and support needs of autistic children above and beyond measures of cognitive ability and autism symptom severity. Adaptive behavior scores provide important information on the needs of autistic people.

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Introduction

Autism research reports often describe study participants who meet diagnostic criteria for autism *and* have cognitive or intelligence quotient (IQ) scores in the average or above average range as "high-functioning" (de Giambattista et al., 2019; Lecavalier, 2014; Szatmari et al., 2003; Venter et al., 1992). It is not uncommon for research designs to incorporate IQ-based functional classifications in their inclusion or exclusion criteria (e.g. Calhoun et al., 2020; Fung et al., 2021; Kim et al., 2023; Pereira et al., 2018; Zajic et al., 2018), and the phrases "low-" and "high-functioning autism" have been used widely in everyday communications as well. This approach is problematic given the consistently reported gap between cognitive and adaptive functioning in autistic individuals (Alvares et al., 2020; Klin et al., 2007; Tamm et al., 2022). Thus, these commonly used classifications of functioning may mask important variability in the relative strengths and difficulties in multiple domains of functioning of autistic individuals and obscure significant needs for support among those with cognitive scores above a given cutoff (Alvares et al., 2020; Wolff et al., 2022).

A similar dichotomy based on cognitive scores is often applied in epidemiologic studies of autism to define co-occurring intellectual disability (ID), with co-occurring ID typically defined to include individuals who both meet diagnostic criteria for autism and have cognitive scores at least two standard deviations (SDs) below the population mean. Many population-based prevalence estimates of co-occurring ID among autistic children are based solely on cognitive scores (Fombonne et al., 2022; Maenner et al., 2020; Van Naarden Braun et al., 2015; Wiggins et al., 2022; Zeidan et al., 2022) despite clinical diagnostic criteria for ID explicitly requiring consideration of adaptive performance as well (Tassé et al., 2016). This study examines classifications of functioning and the relationship between cognitive development and adaptive behavior (AB) in children with autism in a large, well-characterized, community-based sample of preschool-aged children and aims to replicate and build upon relevant findings from previous studies by examining variability in AB scores across the spectrum of cognitive abilities. In doing so, this study provides a critical examination of the use of the terms "low-" and "high-functioning" and "co-occurring ID" based solely on IQ in the autism literature.

Marked variation in AB among individuals with autism has been well-documented across study samples and data sources, age ranges (preschool to adulthood), and even in individuals considered "high-functioning" based on IQ (Alvares et al., 2020; Bölte & Poustka, 2002; Doobay et al., 2014; Fombonne & Zuckerman, 2022; Freeman et al., 1988; Kanne et al., 2011; Klin et al., 2007; Matthews et al., 2015; Paul et al., 2014; Tamm et al., 2022). Several studies looking at domain scores from the Vineland Adaptive Behavior Scales (VABS), one of the most commonly used instruments for measuring AB in autistic individuals, have found evidence of an "autism profile," a characteristic pattern in VABS domain scores with relative strengths in daily living skills, intermediate scores in communication, and greatest delays in socialization (Bölte & Poustka, 2002; Carter et al., 1998; Perry et al., 2009; Venter et al., 1992; Yang et al., 2016), although others have not found this same pattern in their samples (Perry et al., 2009; Ray-Subramanian et al., 2011; Tamm et al., 2022; Tillmann et al., 2019; Yang et al., 2016).

In general, AB scores are positively correlated with cognitive ability (Alvares et al., 2020; Hodge et al., 2021; Kanne et al., 2011; Pathak et al., 2019; Perry et al., 2009; Ray-Subramanian et al., 2011), and the literature suggests this relationship may be stronger in children with autism with lower cognitive scores (Liss et al., 2001; Tillmann et al., 2019; Yang et al., 2016) than in those with higher cognitive scores. Despite this significant association, children with autism commonly exhibit discrepancies between cognitive ability and AB scores (Charman et al., 2011; Klin et al., 2007; Kraper et al., 2017; Tamm et al., 2022; Vig & Jedrysek, 1995), and these gaps have been shown to be larger in individuals with higher cognitive scores and to widen with increasing age (Alvares et al., 2020; Bradshaw et al., 2019; Kanne et al., 2011; Pathak et al., 2019). Large discrepancies in which cognitive scores surpass AB scores have been documented in autistic individuals and IQs above the ID threshold; conversely, in children with autism and cognitive scores two or more SDs below the mean, AB scores are often similar to or exceed IQ (Alvares et al., 2020; Bradshaw et al., 2019; Bölte & Poustka, 2002; Matthews et al., 2015; Pathak et al., 2019; Yang et al., 2016). One longitudinal study found that toddlers with autism with average or above IQs were much more likely than typically developing peers to have below average AB socialization and daily living skills, with IQ/AB discrepancies appearing early and growing with age (Bradshaw et al., 2019). These findings suggest that children with autism may have greater AB strengths and/or delays than would be expected based on their IQ, and these discrepancies may merit consideration in research on functioning of autistic children.

AB scores are also generally inversely associated, albeit weakly or non-significantly, with autism symptom severity as measured by Autism Diagnostic Observation Schedule (ADOS) Calibrated Severity Scores (CSSs) and the Autism Diagnostic Interview–Revised (ADI-R) (Franchini et al., 2018; Hill et al., 2015; Hodge et al., 2021; McDonald et al., 2017; Pathak et al., 2019; Yang et al., 2016). Previous investigators have theorized that IQ/AB discrepancy patterns could be a result of cognitive ability limiting attainment of AB skills in children with lower IQs, while autism symptom severity might attenuate AB skill development in children with higher IQs (Liss et al.,

2001; McDonald et al., 2017; Yang et al., 2016). However, while both cognitive ability and autism symptom severity are associated with adaptive functioning, neither appears to fully explain variation in adaptive functioning, suggesting that AB scores capture unique variation in functioning and may be useful for characterizing support needs and variations in the autism phenotype (Hill et al., 2015; Hodge et al., 2021; Perry et al., 2009; Ray-Subramanian et al., 2011).

Here, we describe AB scores of participants in a large, community-based, epidemiological study of diverse, rigorously characterized 2- to 5-year-old children with autism and examine the association between AB and cognitive development as well as sociodemographic characteristics, autism severity, and behavioral and emotional health. We hypothesize that:

1. There will be significant AB delays in children with autism, even among those with cognitive scores above the ID threshold of two SDs below the mean, with the greatest limitations in socialization, followed by communication, and then daily living skills and motor skills (i.e. the "autism profile").
2. While cognitive scores and autism symptom severity will explain significant variation in AB scores, a large amount of variation in AB scores in children with autism will remain unexplained by these and other factors associated with AB scores, including sociodemographic characteristics and behavioral and emotional health.
3. Children with autism with cognitive scores above the threshold for ID will exhibit significant discrepancies between cognitive and AB scores, with AB scores significantly lower than cognitive scores, while among children with cognitive scores below the ID threshold, AB scores will be similar to or exceed cognitive scores.

Methods

Study sample

The Study to Explore Early Development (SEED) identified children with autism aged 30–68 months by end of data collection through multiple sources, including health, education, early intervention, and other service program settings at study sites within areas of six states (California, Colorado, Georgia, Maryland, North Carolina, Pennsylvania) (Wiggins et al., 2021). Caregivers were asked questions about sociodemographic characteristics, medical history, services and treatments, and other information via telephone interview and mailed questionnaires; caregivers also filled out various standard instruments such as the Child Behavior Checklist (CBCL) (Achenbach, 1992). All participating

families completed the Social Communication Questionnaire (SCQ) (Rutter et al., 2003a). Children who had an autism spectrum disorder (ASD) diagnosis upon enrollment or had ASD risk noted on the SCQ were evaluated with the Mullen Scales of Early Learning (MSEL) (Mullen, 1995) and ADOS (Lord et al., 1999, 2012), and mothers completed the Vineland Adaptive Behavior Scale–Second Edition (VABS-II) (Sparrow et al., 2005) and ADI-R (Lord et al., 1994; Rutter et al., 2003b). Children who did not meet these criteria were evaluated only with the MSEL; if ASD behaviors were witnessed during MSEL administration, the family was asked to also complete the ADOS, ADI-R, and VABS.

Final classification was based on diagnostic criteria for ASD from results of a comprehensive developmental evaluation, including the ADOS and ADI-R, performed by clinicians with established research reliability. Clinical judgment was also considered for children with an early learning age equivalent <24 months who met SEED ASD criteria on the ADOS and ADI-R since the ADI-R is not appropriate for children in this mental age range (Schendel et al., 2012; Wiggins et al., 2015). This population-based ascertainment protocol supported the identification and rigorous assessment of children with autism with and without a previous diagnosis from clinical and community-based sources. Written informed consent was obtained from all participating families. For this research, analysis was restricted to children in the SEED ASD group ($n = 1480$). Sociodemographic characteristics of the sample are provided in Table 1.

Measures

VABS-II: survey interview form. The VABS-II (Sparrow et al., 2005) is a semi-structured interview designed to measure a child's current behavior with an item coding system ranging from "never or rarely performed without help" to "consistently performed without help." The VABS-II was administered in person to the participating child's caregiver. VABS domains include communication, socialization, daily living skills, and in children less than 6 years of age, motor skills, and an overall Adaptive Behavior Composite score can be derived from domain scores. For the composite and all domain scores, standard scores with mean 100, standard deviation 15, and higher scores indicating higher adaptive functioning were used in analyses. VABS-II scores were available for 1458 (98.5%) children with ASD.

MSEL. The MSEL (Mullen, 1995) measures early learning ability in young children up to 68 months of age. In SEED, the MSEL was administered at the in-person assessment, and four subscales on the MSEL—fine motor, visual reception, expressive language, and receptive language—were collected. Subscale *T*-scores have a mean of

Table 1. Child, maternal, and sociodemographic characteristics, overall and stratified by cognitive ability, of children with autism spectrum disorder, Study to Explore Early Development phases one (birth years 2003–2006) and two (birth years 2008–2011).

Demographic characteristic n (%)	Early Learning Composite[a]		
	Overall (n = 1480)	>70 (n = 530)	≤70 (n = 930)
Child age at assessment (years)			
Mean (standard deviation)	4.7 (0.7)	4.7 (0.7)	4.8 (0.7)
Range	2.4–5.8	2.4–5.8	2.6–5.8
Missing	3 (0.2)	0 (0.0)	0 (0.0)
Child sex			
Male	1208 (81.6)	437 (82.5)	752 (80.9)
Female	272 (18.4)	93 (17.6)	178 (19.1)
Child race and ethnicity			
Non-Hispanic White	647 (43.7)	308 (58.1)	329 (35.4)
Non-Hispanic Black	309 (20.9)	57 (10.8)	248 (26.7)
Hispanic	273 (18.5)	79 (14.9)	192 (20.7)
Non-Hispanic other and multiracial	249 (16.8)	86 (16.2)	159 (17.1)
Missing	2 (0.1)	0 (0.0)	2 (0.2)
Child birth order			
Firstborn	702 (47.4)	284 (53.6)	410 (44.1)
Second or later born	713 (48.2)	228 (43.0)	473 (50.9)
Missing	65 (4.4)	18 (3.4)	47 (5.1)
Current income, relative to poverty threshold			
≤100%	133 (9.0)	32 (6.0)	100 (10.8)
101%–200%	277 (18.7)	63 (11.9)	207 (22.3)
201%–299%	234 (15.8)	80 (15.1)	151 (16.2)
300%–399%	220 (14.9)	100 (18.9)	119 (12.8)
≥400%	561 (37.9)	246 (46.4)	309 (33.2)
Missing	55 (3.7)	9 (1.7)	44 (4.7)
Maternal education			
Less than high school	63 (4.3)	10 (1.9)	53 (5.7)
High school graduate	152 (10.3)	33 (6.2)	115 (12.4)
Some college	467 (31.6)	152 (28.7)	306 (32.9)
College graduate	467 (31.6)	176 (33.2)	287 (30.9)
Advanced degree	317 (21.4)	155 (29.3)	160 (17.2)
Missing	14 (1.0)	4 (0.8)	9 (1.0)
Maternal age at enrollment (years)			
Mean (standard deviation)	35.6 (5.6)	36.1 (5.2)	35.3 (5.7)
Range	20-52	21-52	20-52
Mother born in the United States			
Yes	1132 (76.5)	422 (79.6)	694 (74.6)
No	336 (22.7)	105 (19.8)	228 (24.5)
Missing	12 (0.8)	3 (0.6)	8 (1.0)
Primary language spoken at home			
English	1296 (87.6)	492 (92.8)	787 (84.6)
Spanish	78 (5.3)	12 (2.3)	65 (7.0)
Other	93 (6.3)	23 (4.3)	69 (7.4)
Missing	13 (0.9)	3 (0.6)	9 (1.0)
Study site			
California	235 (15.9)	91 (17.2)	139 (15.0)
Colorado	275 (18.6)	122 (23.0)	149 (16.0)
Georgia	285 (19.3)	83 (15.7)	202 (21.7)
Maryland	254 (17.2)	75 (14.2)	179 (19.3)
North Carolina	226 (15.3)	84 (15.9)	137 (14.7)
Pennsylvania	205 (13.9)	75 (14.2)	124 (13.3)

Percentages may not add up to 100 due to rounding.
[a]MSEL Early Learning Composite scores were available for 1460 (98.6%) children with autism spectrum disorder.

50 and standard deviation of 10, and the Early Learning Composite (ELC) is an overall measure of cognitive functioning with a standard score mean of 100 and standard deviation of 15; higher scores on the MSEL indicate higher cognitive ability. This research focused on the MSEL ELC and, to improve precision in the lower range in the regression analysis, a composite raw score was calculated by summing the raw scores from the four subscales. To avoid underestimating early learning ability in children with language delays that are likely present in this preschool-aged sample but may not persist over time or with intervention, we also performed supplemental analyses using the visual reception subscale raw score as a proxy for non-verbal cognitive ability, which has been shown to be more stable in young children with autism than verbal IQ due to the rapid language development evident in verbal scores measured in longitudinal studies of children with autism (Akshoomoff, 2006; Chawarska et al., 2009; Ellis Weismer et al., 2021; Howlin et al., 2004). For analyses stratified by cognitive ability, an ELC threshold of 70 (i.e. two SD below the mean) was used to stratify the sample by whether they would be considered "high-functioning" or "low-functioning" in the literature. MSEL ELC scores were available for 1460 (98.6%) children with ASD. We also conducted a sensitivity analysis using developmental quotients (DQs) and stratifying by a DQ threshold of 70. An overall DQ was calculated by averaging the age equivalent scores from the four subscales, dividing by the child's age in months at administration of the MSEL, and multiplying by 100; a visual reception DQ was also calculated from the visual reception age equivalent. These results are available in the Supplemental Materials (Supplementary Tables S3 and S4).

ADOS. The ADOS (Second Edition; Lord et al., 1999, 2012) is a semi-structured diagnostic child observation that captures social affect and stereotyped behavior and restricted interests in four alternative modules: 1—preverbal/single words, 2—phrase speech/non-fluent, 3—fluent speech (child/adolescent), and 4—fluent speech (adolescent/adult). In SEED, module 1, 2, or 3 was administered at the in-person assessment depending on the expressive language abilities of the child. For this research, ADOS CSSs were used to measure autism symptom severity, with higher CSSs indicating higher severity (Gotham et al., 2009).

Childhood Behavior Checklist/1½–5. The Childhood Behavior Checklist/1½–5 (CBCL/1½–5; Achenbach, 1992) is a parent-administered questionnaire on which a child's caregiver is asked to score behavioral and emotional items on a scale of "not true, as far as you know," "somewhat or sometimes true," and "very true or often true" with higher scores indicating greater problems. The CBCL includes three broad scales: externalizing (which includes attention problems and aggressive behavior), internalizing (which includes anxiety/depression, emotional reactivity, somatic complaints, and withdrawn behaviors), and total problems. CBCL forms were mailed to participating families and filled out by caregivers. *T*-scores with mean 50 and standard deviation 10 were collected for the externalizing, internalizing, and total problems scales with a score of ⩾65 indicating borderline to clinically significant problems and higher scores indicating more problems.

Statistical analysis

For Hypothesis 1, we computed VABS-II summary statistics. Mean scores were compared between VABS-II domains using Friedman tests and post hoc Wilcoxon signed-rank sum tests, overall and within each ELC stratum. To estimate the amount of variance in VABS-II scores explained by cognitive ability and autism symptom severity (Hypothesis 2), hierarchical linear regression analysis was performed. In univariate analysis, child's sex, race, ethnicity, age at administration of the MSEL, and birth order; family's current income relative to the federal poverty level; primary language spoken in the home; maternal age at enrollment, education, and place of birth; and study site were associated with VABS-II scores and included as predictors in Model 1. In subsequent separate steps, different predictors were added to the regression model, including MSEL composite raw score (Model 2); to account for the potential of a curvilinear relationship in which cognitive and adaptive scores were similar in the lower MSEL score range while cognitive development outpaced adaptive scores in the higher MSEL score range, we also included the quadratic term. In Model 3, ADOS CSS was added, followed by the interaction between ADOS CSS and the two cognitive variables (Model 4) in order to explore the potential effect modification of cognitive development on the relationship between adaptive scores and autism severity. Finally, we added CBCL total *T*-score in Model 5 to estimate the additional variance in AB scores explained by overall behavioral and emotional problems (Franchini et al., 2018; Green & Carter, 2014; Kraper et al., 2017). We did not examine externalizing and internalizing problems separately due to significant correlations between these two domains. Nested models were then compared using Wald tests incorporating robust standard errors to determine whether the additional variance explained by a new predictor was significant. Separate regressions were run for the VABS-II composite and each domain score. Regression analysis was limited to complete cases (*n* = 1278 (86.4% of ASD cases) for the composite regression; *n* = 1291 (87.2% of ASD cases) for domain regressions), and we also performed sensitivity analyses using multiple imputation to account for children missing data.

Finally, we described cognitive ability/AB discrepancies among children with ASD with both MSEL ELC and

VABS-II scores recorded (Hypothesis 3), overall and within three age cohorts (2–3 years, 4 years, and 5 years of age). We estimated the percent of children with autism with either, both, or neither ELC or VABS-II score indicating significant challenges and calculated Cohen's kappa coefficients to assess the degree of agreement between the two measures in categorization of functioning (McHugh, 2012). In addition, after converting both ELC and VABS-II scores into SD units rounded to two decimal places, we calculated the magnitude of cognitive ability/AB discrepancies and determined the percent of children within each cognitive stratum with one and two SD discrepancies between the two measures.

Regression analysis was conducted using R Version 4.2.2 (R Core Team, 2022); the R packages "lmtest" (Zeileis & Hothorn, 2002) and "sandwich" (Zeileis, 2004; Zeileis et al., 2020) were used for nested model comparisons. All other statistical analyses were conducted using SAS Version 9.4 (SAS Institute Inc., 2010). The design and implementation of the SEED study have benefited from community stakeholder input from autistic self-advocates, family members, and service providers, although there was no direct community involvement in the reported study.

Results

Hypothesis 1

On average, the SEED ASD group exhibited significant AB delays. As hypothesized, the socialization mean was the lowest and motor skills the highest (1.9 and 1.4 SDs below the mean, respectively). In children with ELC $\leq$ 70, mean VABS-II composite and domain scores were all 1.7 SD or more below the mean. In children with ELC > 70, all mean domain scores were below the population mean of 100, with mean composite, daily living skills, and socialization greater than one SD below the mean.

Contrary to our hypothesis, communication scores were higher than daily living skills on average (Table 2). Between-domain analysis revealed that all mean domain scores significantly differed from one another ($p < 0.0001$ for all comparisons except communication versus daily living skills in children with ELC $\leq$ 70 ($p = 0.03$)), overall and within each ELC stratum. Overall and for children with ELC $\leq$ 70, mean motor skills scores were the highest, followed by communication, daily living skills, and socialization, while in children with ELC > 70, mean communication scores were the highest, followed by motor skills, daily living skills, and finally socialization.

Hypothesis 2

There was wide variation in VABS-II scores in the overall ASD group and within cognitive strata. VABS-II composite scores ranged from 41 to 114 (3.9 SDs below the mean to 0.9 above the mean) (Table 2). Overall and in children with ELC > 70, communication scores had the largest range of any domain, while in children with ELC $\leq$ 70, daily living skills scores had the largest range; socialization had the smallest range of any domain overall and in both strata (Table 2).

In regression analysis, sociodemographic factors explained between 6.0% (motor skills) to 17.2% (communication) of variance in VABS-II scores (Table 3). Addition of the MSEL composite raw score explained significant and substantial variance in VABS-II scores ($\Delta R^2 = 51.1\%$ for the VABS-II composite score), with domain-specific ΔR^2 values ranging from 58.0% in communication to 31.9% in motor skills scores (Model 2a in Table 3). Addition of the ADOS CSS (Model 3a) explained a significant but small amount of additional variance in the communication ($\Delta R^2 = 0.1\%$), daily living skills ($\Delta R^2 = 0.2\%$), and socialization ($\Delta R^2 = 0.4\%$) domains. Addition of the MSEL/ADOS CSS interaction only explained significant additional variance for motor skills ($\Delta R^2 = 0.5\%$) (Model 4a). Finally, addition of the CBCL total T-score explained significant additional variance for all VABS-II scores, explaining most in socialization ($\Delta R^2 = 4.9\%$) and least in motor skills ($\Delta R^2 = 1.0\%$) (Model 5a). Altogether, these predictors explained 67.2% of the variance in composite AB scores, with most explained variance in VABS-II communication scores ($R^2 = 76.4\%$), followed by daily living skills ($R^2 = 57.3\%$), socialization ($R^2 = 48.9\%$), and motor skills ($R^2 = 39.5\%$) (Table 3).

In children with MSEL ELC $\leq$ 70, the full model explained between 72.1% (communication) and 31.9% (motor skills) of AB score variance (Table 3). The MSEL composite raw score explained significant additional variance for every VABS-II score, ranging from 60.2% for communication to 24.1% for motor skills. The ADOS CSS explained significant additional variance for communication ($\Delta R^2 = 0.1\%$), socialization ($\Delta R^2 = 0.8\%$), and motor skills ($\Delta R^2 = 0.4\%$), while the additional variance explained by the MSEL/ADOS CSS interaction was not significant for any score. CBCL total T-score explained significant additional variance for the composite and every domain, ranging from 3.9% for socialization to 0.6% for motor skills.

In comparison, in children with ELC > 70, these predictors explained less of the VABS-II score variance with R^2 values ranging from 39.0% for communication to 16.9% for motor skills (Table 3). The MSEL composite raw score explained significant additional variance for every VABS-II score, ranging from 13.4% for communication to 1.2% for socialization. Addition of the ADOS CSS explained significant additional variance in daily living skills ($\Delta R^2 = 0.9\%$) and motor skills ($\Delta R^2 = 1.1\%$), while the MSEL/ADOS CSS interaction did not explain significant additional variance for any VABS-II score. CBCL total T-scores explained significant additional variance for

Table 2. Summary statistics for selected instruments, overall and stratified by cognitive ability, for children with autism spectrum disorder, Study to Explore Early Development phases one (birth years 2003–2006) and two (birth years 2008–2011).

Variable	Overall			MSEL Early Learning Composite					
				≤70			>70		
	Mean	Standard deviation	Range	Mean	Standard deviation	Range	Mean	Standard deviation	Range
VABS-II									
Composite	73.2	13.3	41–114	67.3	11.2	41–98	83.6	9.9	47–114
Communication	77.8	17.7	36–131	69.7	15.5	36–108	92.1	11.1	40–131
Daily living skills	74.7	15.1	34–121	69.0	14.0	34–109	84.8	11.3	48–121
Socialization	72.1	12.6	44–112	67.6	11.2	44–105	80.0	11.1	49–112
Motor skills	79.7	13.4	49–124	74.9	12.0	49–117	88.0	11.7	49–124
MSEL									
Early learning composite	66.6	19.6	49–132	53.6	6.3	49–70	89.3	13.3	71–132
Composite raw score	131.0	39.7	14–197	109.9	32.8	14–173	167.8	17.8	94–197
Visual reception *T*-score	34.7	15.5	20–80	25.5	8.8	20–58	51.0	10.5	20–80
Visual reception raw score	37.4	10.2	0–50	32.3	9.2	0–50	46.3	4.0	27–50
Fine motor *T*-score	29.6	11.7	20–80	23.0	6.1	20–52	41.1	10.3	20–80
Fine motor raw score	33.8	8.8	6–49	29.6	7.4	6–47	41.2	5.6	23–49
Receptive language *T*-score	30.2	12.9	20–73	22.0	4.4	20–51	44.5	10.2	20–73
Receptive language raw score	30.5	11.5	0–48	24.5	9.4	0–45	41.1	5.6	20–48
Expressive language *T*-score	29.1	11.4	20–74	22.5	5.1	20–49	40.8	9.8	20–74
Expressive language raw score	29.2	11.5	3–50	23.5	9.9	3–44	39.3	5.7	20–50
ADOS									
Calibrated severity score	7.3	1.6	4–10	7.6	1.6	4–10	6.7	1.6	4–10
CBCL									
Total *T*-score	62.9	11.1	32–98	63.4	10.5	36–98	62.0	12.1	32–94
Externalizing *T*-score	59.7	11.3	28–97	60.0	10.7	35–97	59.2	12.4	28–92
Internalizing *T*-score	62.6	9.6	29–97	62.6	9.2	33–97	62.4	10.4	29–97

MSEL: Mullen Scales of Early Learning; VABS-II: Vineland Adaptive Behavior Scales–Second Edition; ADOS: Autism Diagnostic Observation Schedule; CBCL: Childhood Behavior Checklist.

every VABS-II scores, ranging from 9.0% for socialization to 2.5% for motor skills. For all scores except communication, the additional variance explained by CBCL scores was similar to or greater than that explained by the MSEL composite raw score, particularly in the socialization domain in which the CBCL total *T*-score explained 9.0% significant additional variance compared to only 1.2% significant additional variance explained by the MSEL composite raw score (Table 3).

Analysis of multiply imputed data resulted in similar R^2 and ΔR^2 values. In addition, patterns were generally similar when the visual reception raw score was used as our measure of cognitive ability, except that, generally, the visual reception raw score explained a smaller amount of additional variance in VABS-II scores than the composite raw score (see Supplementary Table S2).

Hypothesis 3

Figure 1 shows the wide range of composite AB scores among children with ELC scores below and above the two

SD below the mean cutoff (Figure 1). Cohen's kappa coefficients indicated weak (composite (0.45)) to minimal (communication (0.36), daily living skills (0.37), socialization (0.38), and motor skills (0.26)) agreement between MSEL ELC and VABS-II scores regarding whether or not both scores fell above or below the two SD cutoff for significant functional impairment or delay. In our sample, 25.3% of children had ELC ≤ 70 but VABS-II composite scores indicating no significant adaptive delays, while 3.3% and 8.1% had ELC > 70 but VABS-II composite and socialization scores, respectively, indicating significant delay (Figure 1 and Supplementary Table S5). Over a third (39.7%) of children with ASD and ELC ≤ 70 did not have a composite AB score indicating significant adaptive delay (Figure 1, Quadrant I). In addition, among children with ELC > 70, 9.0% and 22.2% had VABS-II composite and socialization scores, respectively, indicating significant adaptive delay (Figure 1, Quadrant IV; Supplementary Table S5). Similar results for other VABS-II domain scores are provided in Supplementary Table S5. Patterns were similar when the relationship between VABS-II scores and

Table 3. Results of linear regression modeling Vineland Adaptive Behavior Scales–Second Edition scores in children with autism spectrum disorder, Study to Explore Early Development phases one (birth years 2003–2006) and two (birth years 2008–2011).

	Model	Variables added	VABS-II score									
			Composite		Communication		Daily living skills		Socialization		Motor skills	
			R² (%)	ΔR² (%)	R² (%)	ΔR² (%)	R² (%)	ΔR² (%)	R² (%)	ΔR² (%)	R² (%)	ΔR² (%)
Overall		n	1278		1291		1291		1291		1291	
	1a	Demographics[a]	13.2	NA	17.2	NA	14.1	NA	10.5	NA	6.0	NA
	2a	+MSEL composite raw score[b]	64.3	51.1***	75.2	58.0***	54.8	40.7***	43.5	33.0***	37.9	31.9***
	3a	+ADOS calibrated severity score	64.4	0.0	75.3	0.1*	55.0	0.2*	43.9	0.4**	37.9	0.0
	4a	+Interaction[c]	64.4	0.0	75.3	0.0	55.0	0.0	44.0	0.2	38.4	0.5**
	5a	+CBCL total T-score	67.2	2.8***	76.4	1.1***	57.3	2.3***	48.9	4.9***	39.5	1.0***
MSEL Early Learning Composite ≤70		n	794		802		802		802		802	
	1b	Demographics[a]	10.4	NA	11.0	NA	15.7	NA	9.2	NA	6.6	NA
	2b	+MSEL composite raw score[b]	61.4	50.9***	71.2	60.2***	53.8	38.1***	42.9	33.8***	30.8	24.1***
	3b	+ADOS calibrated severity score	61.4	0.0	71.4	0.1*	53.8	0.0	43.7	0.8**	31.2	0.4*
	4b	+Interaction[c]	61.5	0.1	71.4	0.1	53.9	0.1	43.8	0.1	31.3	0.0
	5b	+CBCL total T-score	63.6	2.2***	72.1	0.7***	56.1	2.1***	47.7	3.9***	31.9	0.6*
>70		n	484		489		489		489		489	
	1c	Demographics[a]	13.3	NA	20.0	NA	11.4	NA	11.2	NA	9.6	NA
	2c	+MSEL composite raw score[b]	20.1	6.9***	33.4	13.4***	14.9	3.5**	12.3	1.2*	13.0	3.4**
	3c	+ADOS calibrated severity score	20.4	0.3	33.8	0.4	15.7	0.9*	12.5	0.1	14.0	1.1*
	4c	+Interaction[c]	20.6	0.1	33.8	0.0	15.9	0.2	13.0	0.6	14.4	0.4
	5c	+CBCL total T-score	28.7	8.1***	39.0	5.2***	20.2	4.3***	22.0	9.0***	16.9	2.5**

VABS-II: Vineland Adaptive Behavior Scales–Second Edition; NA: not applicable; MSEL: Mullen Scales of Early Learning; ADOS: Autism Diagnostic Observation Schedule–Second Edition; CBCL: Childhood Behavior Checklist.

[a]Demographic variables: child's sex, child's race and ethnicity, child's age at administration of MSEL, child's birth order, household income, primary language spoken in the home, maternal age at enrollment (and the quadratic term), maternal education, mother's place of birth, and study site.

[b]Includes MSEL composite raw score and its quadratic term.

[c]Interaction between MSEL variables and ADOS calibrated severity score.

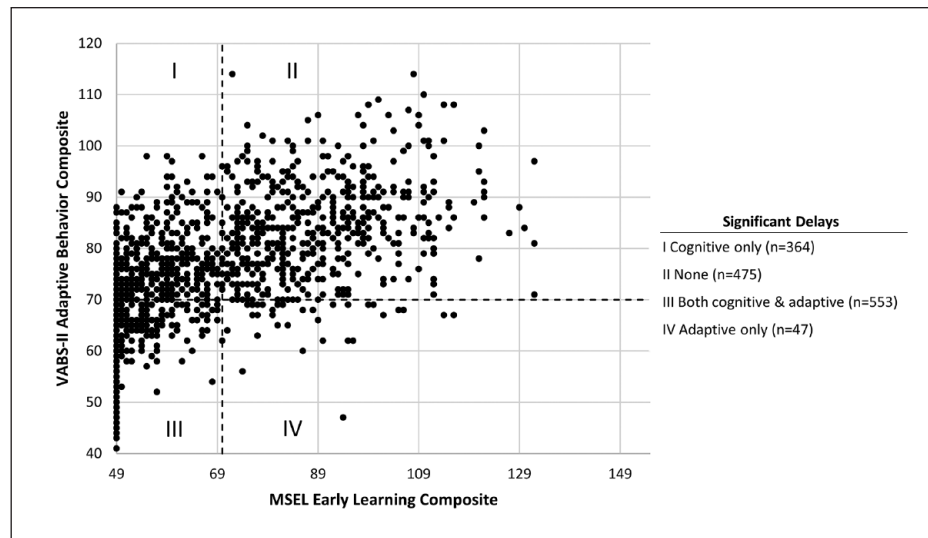*$p < 0.05$; **$p < 0.01$; ***$p < 0.0001$.

Figure 1. Discrepant (I, IV) and concordant (II, III) categorizations of functioning based on cognitive development as measured by the Mullen Scales of Early Learning Composite and/or adaptive behavior scores as measured by the Vineland Adaptive Behavior Scales–Second Edition Composite in children with autism spectrum disorder, Study to Explore Early Development phases one (birth years 2003–2006) and two (birth years 2008–2011). Dashed lines indicate two standard deviations below the mean on the VABS-II (horizontal line) and MSEL (vertical line), the cutoff typically used to indicate significant impairment or delay.
VABS-II: Vineland Adaptive Behavior Scales–Second Edition; ABC: Adaptive Behavior Composite; MSEL: Mullen Scales of Early Learning; ELC: Early Learning Composite.

the MSEL visual reception *T*-score was examined, but a smaller percentage of children had cognitive scores indicating significant delay when measured by the visual reception *T*-score rather than ELC (49.5% vs 63.7%) (see Supplementary Figure S1).

Consistent with our third hypothesis, we found discrepancies between MSEL ELC and VABS-II scores. Most children with ELC > 70 had cognitive scores exceeding their VABS-II scores (62.1% for the composite). The largest discrepancies were seen in the socialization domain, with 36.2% of children having ELC > AB with at least a one SD discrepancy and 12.3% having a two SD or more discrepancy between the two scores (Figure 2). Discrepancies were also large for other domains (Figure 2). In children with ELC ≤ 70, adaptive scores often exceeded ELC (91.9% for the composite) and the proportion with AB scores at least one SD greater than their ELC ranged from 73.8% for motor skills to 43.1% for socialization (Figure 3).

When stratified by age, the proportion of children with a two SD or more discrepancy between their ELC and VABS-II scores decreased with increasing age (Figures 2 and 3), with this trend generally more pronounced in children with ELC > 70. There were substantial proportions of children with at least a one or more SD difference in all age cohorts.

Discussion

Our findings of wide variation in adaptive functioning of preschool children with autism may have important

implications for identifying service needs and targets of interventions, tracking short- and long-term outcomes, and describing functioning in future epidemiologic and clinical studies of ASD. With respect to the VABS "autism profile," we confirmed that children with ASD have significant socialization delays and, in comparison to other domains, relatively fewer motor delays, although scores in this domain still indicated meaningful limitations. In contrast to other studies (e.g. Bölte & Poustka, 2002; Perry et al., 2009; Yang et al., 2016), we found that communication scores exceeded daily living skills scores on average in our sample. These findings suggest that, while evidence for a characteristic "autism profile" is mixed, AB domain scores may be useful for characterizing more precise profiles of strengths and support needs in research examining phenotypic variation in children with autism.

Consistent with our hypothesis, neither cognitive development as measured by MSEL raw scores nor autism severity fully explained variation in AB scores in this sample, particularly in the socialization and motor skill domains and in children with cognitive scores above the ID threshold, suggesting that AB scores capture important variation in the ASD phenotype otherwise missed by these other measures. As in past research in children with ASD (e.g. Liss et al., 2001; Pathak et al., 2019; Yang et al., 2016), we found a strong positive relationship between cognitive ability and AB scores, especially for the communication domain, although, in the overall ASD group, cognitive scores explained less than half of the variation in daily living skills, socialization, and motor skills and even
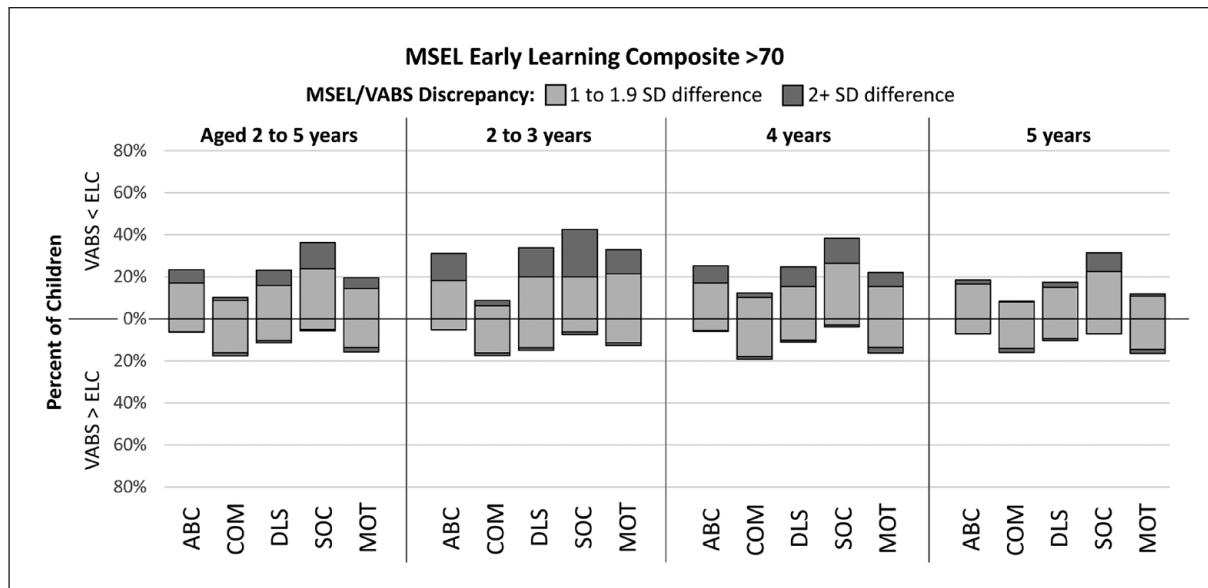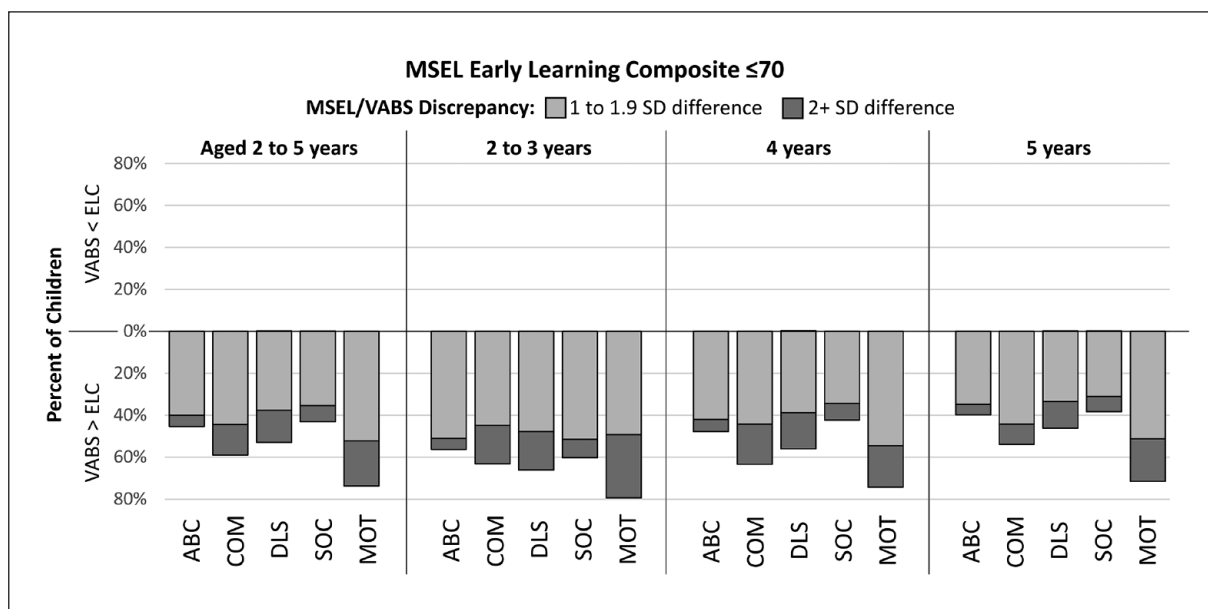
Figure 2. Discrepancies of one or more SD between cognitive development as measured by the Mullen Scales of Early Learning (MSEL) Composite (ELC) and Vineland Adaptive Behavior Scales–Second Edition Composite (ABC), communication (COM), daily living skills (DLS), socialization (SOC), and motor skills (MOT) scores among children with autism spectrum disorder and MSEL ELC > 70, Study to Explore Early Development phases one (birth years 2003–2006) and two (birth years 2008–2011).
VABS: Vineland Adaptive Behavior Scales–Second Edition; ELC: Early Learning Composite; SD: standard deviation; ABC: Adaptive Behavior Composite.

Figure 3. Discrepancies of one or more SD between cognitive development as measured by the Mullen Scales of Early Learning (MSEL) Composite (ELC) and Vineland Adaptive Behavior Scales–Second Edition Composite (ABC), communication (COM), daily living skills (DLS), socialization (SOC), and motor skills (MOT) scores among children with autism spectrum disorder and MSEL ELC ≤ 70, Study to Explore Early Development phases one (birth years 2003–2006) and two (birth years 2008–2011).
VABS: Vineland Adaptive Behavior Scales–Second Edition; ELC: Early Learning Composite; SD: standard deviation; ABC: Adaptive Behavior Composite.

less within each cognitive stratum, explaining less than 14% of variation in any VABS-II score in children with ELC > 70. Severity of autism symptoms explained little

variance while CBCL scores explained somewhat more, particularly in the socialization domain and in children with ELC > 70, consistent with previous findings

of a significant negative association between adaptive functioning and behavioral and emotional problems (Franchini et al., 2018; Green & Carter, 2014; Kraper et al., 2017).

Finally, as hypothesized, we found that children with autism with cognitive scores indicating no significant cognitive delay exhibited substantial cognitive/AB discrepancies; many of these children had cognitive scores substantially exceeding AB scores, although contrary to our hypothesis, we saw a higher proportion of substantial cognitive/AB discrepancies in children with cognitive scores two or more SDs below the mean. These discrepancy findings are consistent with past research of children across different age groups (e.g. Alvares et al., 2020; Pathak et al., 2019; Tillmann et al., 2019). Together these results suggest that using cognitive scores, autism symptom severity, and behavioral and emotional problems as a proxy for "functioning"—either together or separately—obscures heterogeneity in adaptive functioning among children with autism and that large gaps between cognitive development and AB are apparent in children with autism across the spectrum of cognitive development as early as preschool. These findings may be important when designing treatment plans and interpreting research findings.

Our results did not support the theory that cognitive ability would be the limiting factor in attainment of AB skills in children with lower cognitive ability and autism severity for children with greater cognitive development (Liss et al., 2001; McDonald et al., 2017; Yang et al., 2016), as an interaction term between cognitive score and autism symptom severity did not explain substantial additional variance in AB scores and ADOS CSSs did not explain substantial additional variance in either cognitive stratum. However, previous study samples often included older individuals, and longitudinal studies have found an association between greater autism symptom severity and low or declining trajectories of AB skill attainment as well as widening IQ/AB gaps with increasing age (Bradshaw et al., 2019; Franchini et al., 2018; Green & Carter, 2014). Because our sample was preschool-aged, the relationship between autism symptom severity and adaptive functioning and the influence of symptom severity on the gap between cognitive development and AB scores could strengthen as they age and develop. While we actually saw fewer large cognitive ability/AB discrepancies in 4- and 5-year-old children relative to those less than 4 years of age, previous research has shown that issues related to testability may impact how young children with ASD perform on the MSEL (Akshoomoff, 2006; Courchesne et al., 2019), and therefore, the MSEL may have underestimated the cognitive abilities—especially related to language—of the children in our sample, particularly those under 4 years of age.

Other factors that may contribute to variation in adaptive functioning in children with autism are behavioral or emotional problems and co-occurring physical and mental health conditions. Previous studies have found associations between the two (Franchini et al., 2018; Green & Carter, 2014; Kraper et al., 2017), although others have not (Tillmann et al., 2019). Our results suggest greater behavioral and emotional problems are associated with lower adaptive functioning, particularly in the socialization domain. In our sample, CBCL scores explained greater additional variance in AB scores than did autism severity, and in children with ELC scores indicating no significant cognitive delays, CBCL scores explained similar or greater additional variance than even cognitive scores in every domain except communication.

Strengths and limitations

The large, population-based SEED sample includes children with ASD from diverse racial, ethnic, and socioeconomic backgrounds. Participants all underwent rigorous, research-reliable, professional assessments and had nearly complete data on instruments measuring cognitive development, adaptive functioning, autism symptom severity, and behavioral and emotional health. In addition, recruitment procedures allowed for identification of ASD cases from a variety of community sources that may otherwise be missed when participants are recruited primarily from clinical populations, as is the case for much of the previous literature on adaptive functioning in children with autism. The broad diagnostic net for case identification in SEED also made possible the inclusion of children without a previous ASD diagnosis, mitigating the risk of a sample biased toward children with access to healthcare or intervention services.

One limitation of our study was the lack of a measure of social demands and support, particularly given our finding that sociodemographic characteristics explained variation in adaptive functioning (Constantino et al., 2020; Mandell et al., 2005). Future research could examine the association between AB and measures of social determinants of health, such as access to resources like early diagnosis and early intervention programs and family and neighborhood environment. Future research could also examine domain-specific associations between cognitive ability, autism symptom severity, and behavioral and emotional problems and AB scores.

Another consideration is the restricted age range of children in our sample. It is important to note that ID is not typically diagnosed in preschool-aged children due to the instability of IQ scores in this age group. However, early childhood cognitive scores have been shown to be predictive of adult outcomes, including IQ (Bishop et al., 2015; Magiati et al., 2014; Pickles et al., 2020), and this study provides evidence that the gap between cognitive and AB scores found in previous ASD research emerges early and may have important implications for research on ASD with co-occurring ID and interpretation and of past

research that classified children with autism as "high-" or "low-functioning" based solely on cognitive scores. Finally, this analysis focused on the MSEL and VABS-II, and the findings may not generalize to all measures of cognitive and adaptive development.

Conclusion

Our finding of significant heterogeneity in adaptive functioning and substantial discrepancies between cognitive and AB scores in preschool-aged children with autism suggests that understanding the relationship between AB and cognitive ability could lead to better characterization of the heterogeneity and needs for support of young autistic children. Autistic children, particularly those without significant cognitive delays, may have support needs not reflected by their cognitive ability; likewise, children with cognitive delays may have adaptive strengths and abilities not evident based on their cognitive scores. In our sample, reliance on cognitive scores alone to identify level of functioning among young children with ASD would potentially designate 25.3% of children as "low-functioning" or having co-occurring ID despite having no significant delays in overall adaptive functioning and 3.3% of children as "high-functioning" despite having significant delays in overall adaptive functioning. Moreover, 8.1% of children could be classified as "high-functioning" despite having significant delays in VABS-II socialization scores. In terms of service needs, autistic children who have cognitive scores indicating no significant delays have a range of adaptive ability levels and many may benefit from AB interventions.

These findings support concerns often raised by researchers and autistic advocates that categorical labels of functioning may fail to capture an individual's unique strengths, challenges, and needs for support (Alvares et al., 2020; Bal et al., 2017; Bottema-Beutel et al., 2021). They also suggest that reliance on cognitive scores alone may not be sufficient for describing the epidemiology of co-occurring ID and autism. Cognitive development explained only approximately half of the variation in overall adaptive functioning in our full sample and even less in children without significant cognitive delays. This suggests that assignment of functioning labels based on this measure offers an incomplete and potentially inaccurate picture of the abilities and support needs of autistic children. Future epidemiologic studies and other research focused on children with autism with and without co-occurring cognitive delays could consider incorporating measures of adaptive functioning and examining domain-specific patterns of strengths and weaknesses to better characterize functional abilities.

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Supplemental material

Supplemental material for this article is available online.

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