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Association between severity of behavioral phenotype and comorbid attention deficit hyperactivity disorder symptoms in children with autism spectrum disorders

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Abstract

Autism spectrum disorder and attention deficit hyperactivity disorder are neurodevelopmental disorders that cannot be codiagnosed under existing diagnostic guidelines (*Diagnostic and Statistical Manual of the American Psychiatric Association*, 4th ed., text rev.). However, reports are emerging that attention deficit hyperactivity disorder is sometimes comorbid with autism spectrum disorder. In the current study, we examined rates of parent-reported clinically significant symptoms of attention deficit hyperactivity disorder in school-aged children (4–8 years) with autism spectrum disorder, most of whom were first enrolled in our research protocols as toddlers. Results revealed that children with autism spectrum disorder and attention deficit hyperactivity disorder had lower cognitive functioning, more severe social impairment, and greater delays in adaptive functioning than children with autism spectrum disorder only. Implications for clinical practice include the need to assess for attention deficit hyperactivity disorder symptoms at an early age in children diagnosed with autism spectrum disorder. Research is needed to determine efficacious interventions for young children with autism spectrum disorder with comorbid attention deficit hyperactivity disorder to optimize outcomes.

Keywords

attention deficit hyperactivity disorder; autism; comorbidity; symptom severity

Introduction

Autism spectrum disorder (ASD) and attention deficit hyperactivity disorder (ADHD) are neurodevelopmental disorders with onset of symptoms in early childhood. ASD is characterized by impairments in communication and social reciprocity and stereotypic and/or repetitive behaviors, with symptoms presenting by the age of 3 years (American Psychiatric Association (APA), 2000). ADHD, the most common psychiatric disorder diagnosed in childhood (Wilens et al., 2002), is characterized by symptoms of inattention, impulsivity, and/or hyperactivity beyond what would be expected for developmental level and presents before the age of 7 years (Wilens et al., 2002).

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There is overlap in the clinical presentation of ASD and ADHD. Both disorders include communication problems, restricted behaviors, and problems with attention (Hattori et al., 2006), and are more prevalent in boys than girls, with ratios of 4:1 in ASD (Centers for Disease Control (CDC), 2012) and approximately 3:1 in ADHD (CDC, 2010). Moreover, epidemiological studies have identified increasing prevalence rates of ASD (CDC, 2012) and ADHD (CDC, 2010) over the past decade. Symptoms associated with both disorders cause significant behavioral, social, and adaptive problems across home, school, and community settings (Rich et al., 2009). There is preliminary evidence that when ADHD is comorbid with ASD, the risk for increased severity of psychosocial problems increases (Holtmann et al., 2007; Yerys et al., 2009). More externalizing, internalizing, and social problems (Holtmann et al., 2007), as well as more impaired adaptive functioning, and more autistic traits and maladaptive behaviors (Yerys et al., 2009) have been reported in children with ASD comorbid with ADHD than children with ASD only.

Although symptoms of ADHD are often clinically noted in children with ASD (Mayes et al., 2012), the *Diagnostic and Statistical Manual of the American Psychiatric Association* (4th ed., text rev.; *DSM-IV-TR*; APA, 2000) prohibits a dual diagnosis of ADHD and ASD (APA, 2000). However, a growing number of researchers are reporting symptoms meeting *DSM-IV-TR* criteria for ADHD in a substantial proportion of children diagnosed with ASD, and there is increasing evidence that these two disorders may indeed co-occur (Brereton et al., 2006; Gadow et al., 2006; Goldstein and Schwebach, 2004; Holtmann et al., 2007; Lee and Ousley, 2006; Leyfer et al., 2006; Sinzig et al., 2009; Yerys et al., 2009). A significant percentage of children with ASD seeking services at clinical centers present with comorbid symptoms of ADHD, with rates of comorbidity ranging between 37% (Gadow et al., 2006) and 85% (Lee and Ousley, 2006) across studies conducted in the United States and Europe.

Little is known, however, about ADHD comorbidity rates in nonclinical populations of children with ASD. Only one study using a community sample of children was identified in the extant literature. Leyfer and colleagues (2006) found that ADHD was the third most common psychiatric disorder identified in a community sample of children with ASD, ranging in age from 5 to 17 years, with 31% of the sample meeting full diagnostic criteria for ADHD and another 24% of the sample identified with subsyndromal ADHD symptoms. This estimate is lower than reported rates of comorbid ASD and ADHD in clinic samples.

A major need in the field of autism research is to better understand how often clinically significant ADHD symptoms are comorbid with ASD in nonclinical populations, and whether the comorbidity of ADHD with ASD is related to differences in other aspects of behavioral phenotype. We address this need in the current article. We also examine the ADHD comorbidity with ASD in a more restricted age range than has been previously studied (e.g. Gadow et al.'s (2006) sample of 3- to 12-year-olds and Lee and Ousley's (2006) sample of children (mean age = 8.7 years) and adolescents (mean age = 15.3 years)), providing clarity about phenotype and ADHD risk in the early school-age years. Greater understanding of how comorbid disorders relate to other aspects of behavioral phenotype in young children with ASD could inform clinical and educational assessments and intervention decisions that could ultimately impact children's psychosocial outcomes.

In the current study, we first examined rates of parent-reported clinically significant symptoms of ADHD in school-aged children with ASD. We focused on a non-clinical sample in order to avoid overestimating the rate of ADHD comorbidity with ASD, as clinical samples may be biased toward more severely impaired children. We tested the hypothesis that children with ASD and comorbid ADHD would exhibit a more severe behavioral phenotype than those with ASD only. Specifically, we hypothesized that the comorbid group would have lower cognitive functioning, greater delays in adaptive functioning, higher rates of internalizing problems, and more severe social impairment than children with ASD only when these groups were of similar age and did not differ in recruitment source.

Method

This study was approved by the Johns Hopkins University Institutional Review Board before the collection of data, and all families gave written consent for the participation of their children.

Participants

Participants included 162 children (100 males), ages 4–8 years (M = 5.6) enrolled in a prospective, longitudinal study of child development, consisting of (a) full biological younger siblings of children with autism who had been followed in our research program since at least 14 months of age (with ASD n = 27 and without ASD n = 75); (b) children diagnosed with ASD (n = 35), including the older probands of younger siblings participating in the study (n = 8), and children who had participated in early intervention studies (n = 27) at the Kennedy Krieger Institute (KKI); (c) low risk controls with no known family history of ASD (n = 12); and (d) children with a history of specific language delay who had been followed in our research program since age 18 or 24 months (n = 13). Children without ASD, including the 13 children with non-ASD language delays, were included in this study to provide a reference point for the level of functioning and rates of impairment in the ASD groups.

ASD and ADHD classification—Grouping of participants was strategically planned to address the research questions. Children were first divided into ASD and non-ASD groups. They were then further categorized according to parent-reported symptoms of ADHD on the Hyperactivity and Attention Problems subscales of the Behavioral Assessment System for Children–Second Edition (BASC-2; Reynolds and Kamphaus, 2004).

ASD classification—Master's- and doctoral-level clinical researchers with expertise in autism and child development administered the Autism Diagnostic Observation Schedule–Generic (ADOS; Lord et al., 2002). The ADOS is a semistructured, play-based assessment designed to evaluate social communicative behaviors characteristic of ASD. Assignment to the ASD group required meeting ADOS algorithm criteria for ASD or autism and having a clinical judgment of ASD based on *DSM-IV-TR* criteria for autism or Pervasive Developmental Disorder–Not Otherwise Specified (PDD-NOS). For our research purposes,

we used the ASD classification without further subtyping the children into autism or PDD-NOS categories. There were 62 children with ASD.

ADHD classification—The criterion for receiving an ADHD classification was based on parent ratings on the Hyperactivity and Attention Problems subscales of the parent version of the BASC-2 (Reynolds and Kamphaus, 2004). According to the BASC-2 manual, these two subscales were specifically designed to identify the core symptoms of ADHD as specified in the *DSM-IV-TR* (i.e. hyperactivity, impulsivity, and inattention). Children were classified as ADHD-positive if they scored at or above the BASC-2 clinical threshold (≥70) on either the Hyperactivity or Attention Problems subscales. There were nine non-ASD children who scored above the clinical threshold on at least one of the two ADHD subscales. The rate of parent-reported symptoms in the children without ASD in this study (9%) is similar to the rate reported for the general population of American children (9.5%; CDC, 2010). We elected not to form a separate ADHD group with these nine children because the small sample size would not permit us to draw meaningful conclusions from group comparisons. Thus, these nine children were not included in any of the analyses.

Recruitment and procedure

Participants for this study were recruited through fliers posted at the Kennedy Krieger Institute (KKI), autism advocacy groups, community events, and local autism conferences. Children were assessed either at KKI or at the Massachusetts General Hospital (MGH).

For younger siblings of children with ASD to be eligible, the proband (older sibling) was required to meet diagnostic algorithm criteria for ASD or autism on the ADOS (Lord et al., 2002), diagnostic algorithm criteria for autism on the Autism Diagnostic Interview–Revised (ADI-R; LeCouteur et al., 1995), and have a clinical judgment of ASD by an expert clinical researcher. Eligibility criteria for children in all groups required that they have no history of severe birth trauma or severe head injury and no diagnosis of a genetic anomaly. In addition, the primary language spoken in the home had to be English. The language delayed and low risk controls were required to have a negative family history of major psychiatric disorders. Low risk controls also had no family history of language delay. Family history of ASD, language delay, and major psychiatric disorder was determined through a telephone screening interview with one of the parents (usually the mother).

Annual evaluations were identical at both sites for younger siblings of children with ASD (the only participants assessed at more than one site) and for all groups of children. These evaluations included a comprehensive battery of tests measuring cognitive, language, and social development, including the ADOS. Testers at both sites were master's- and doctoral-level professionals with training in assessing and diagnosing children with ASD and expertise in child development. Testers from the MGH site received training in the testing protocol at the KKI site by expert testers and testers from both the KKI and MGH sites received ADOS training by certified ADOS trainers at KKI. All testers were research reliable in ADOS administration and scoring. There was high tester agreement in final case classification within and across sites (i.e. ASD or no ASD; $\alpha = .87$, $p < .001$). Final case

classification was determined by the second author and principal investigator of the study when there was disagreement.

The study protocol included clinician-administered gold standard diagnostic assessments of ASD, standardized assessments of language and cognitive development, and parent-completed questionnaires of social and adaptive functioning and developmental psychopathology. Data for the current study were taken from the most recent annual assessment for which parents had completed the BASC-2 (Reynolds and Kamphaus, 2004). The sample comprised 41 (25%) 4-year-olds, 47 (29%) 5-year-olds, 31(19%) 6-year-olds, 30 (19%) 7-year-olds, and 13 (8%) 8-year-olds.

Measures

Cognitive functioning—Two instruments were used to assess cognitive functioning. The majority of the participants completed the Stanford-Binet Intelligence Scales, 5th edition (SB-5; Roid, 2003). The SB-5 is a standardized assessment of intellectual functioning, providing a full-scale composite score (FSIQ) comprising five ability factors, providing nonverbal (NV) and verbal (V) domain scores. The standard scores have a mean of 100 and a standard deviation of 15. Ten children with ASD diagnoses who were unable to complete the SB-5 due to low cognitive functioning completed the Mullen Scales of Early Learning (MSEL; Mullen, 1995). In addition to standard scores, the MSEL provides age equivalency scores for four of the five scales: Visual Reception, Fine Motor, Expressive Language, and Receptive Language. Because the MSEL does not provide standard scores for children over the age of 68 months, we used age equivalency scores from the NV domain of the SB-5 and the Visual Reception scale of the MSEL as a common metric of NV cognitive functioning.

Psychopathology—The BASC-2 (Reynolds and Kamphaus, 2004), Parent Edition, Internalizing Problems Composite Scale served as a measure of developmental psychopathology involving mood and anxiety disorders. We selected the Internalizing Composite Scale, which includes the Anxiety, Depression, and Somatization subscales, because the BASC-2 manual (Reynolds and Kamphaus, 2004) reports that it provides a better indicator of mood and anxiety disorders than scores yielded from the constituent individual scales. Internalizing Composite Scale scores between 60 and 69 indicate high risk for psychopathology, and scores ≥ 70 indicate the presence of clinically significant symptoms of psychopathology.

Social functioning and autism-related mannerisms—The Social Responsiveness Scale (SRS; Constantino and Gruber, 2005) served as a measure of social functioning and autism-related mannerisms. The SRS is a standardized parent report questionnaire designed to assess social functioning in children and adolescents with ASD between the ages of 4 and 18 years. The SRS consists of five subscales: Social Awareness (ability to detect social cues from the environment), Social Cognition (ability to cognitively process and interpret social cues), Social Communication (NV and V pragmatic capabilities), Social Motivation (desire to engage in social interaction), and Autistic Mannerisms (stereotypic behaviors and restricted interests), which yield standard T-scores based on age-specific norms. In addition to these five subscales, the SRS yields a composite or Total Social Functioning T-score. T-

scores between 60 and 75 indicate mild to moderate social impairment; T-scores ≥ 76 indicate clinically significant or severe social impairment.

Adaptive behavior—The Vineland Adaptive Behavior Scales, Second edition (VABS-II; Sparrow et al., 2007) is a clinician-administered, semistructured parent interview that we used to obtain parent ratings of children's adaptive functioning across three domains: Communication, Socialization, and Daily Living Skills. The VABS-II provides an indicator of the degree to which daily adaptive skills are impacted over and above what would be expected given IQ level in children with developmental disabilities. The VABS-II standard scores have a mean of 100 and standard deviation of 15. The VABS-II provides the following categorical levels of adaptive functioning: high (≥130), moderately high (115–129), adequate (86–114), moderately low (71–85), and low (≤70).

Results

All statistical analyses were conducted using the Statistical Package for the Social Sciences (SPSS, v. 18). The data analysis was conducted with a final sample of 153 participants (excluding the 9 non-ASD children with parent-reported clinically significant ADHD symptoms), consisting of the following classification groupings: No ASD (n = 91), ASD Only (n = 44), and ASD+ADHD (n = 18). There were two sibling pairs (consisting of one proband and one younger sibling from the same family) in this final sample, violating the assumption of independence. To control for this possible violation, we conducted the data analyses without the two younger siblings and found that the results were no different from when these children were included in the sample. Thus, we included them in the sample. The ASD+ADHD group met the clinical threshold on the following subscales of the BASC-2: Hyperactivity (n = 6; 33%), Attention Problems (n = 6; 33%), and both sub-scales (n = 6; 33%).

Table 1 presents mean age and mean NV cognitive age equivalency by group, along with the following demographic information: gender, ethnicity, and socioeconomic status (SES). There were no significant between-group differences in age, ethnicity, or SES (ps > .05). The mean age for all groups was very similar, and the majority of the sample was Caucasian and belonged to middle to upper SES. As expected, given the gender ratio of ASD (APA, 2000), there were significantly more males than females in the two ASD groups than in the No ASD group (χ^2 = 22.61, p < .001). There were also significantly more males in the ASD +ADHD group than in the ASD Only group (100% and 75%, respectively; Fisher's exact test, p = .025). The mean NV cognitive age equivalency score for the No ASD group was significantly higher than that of the two ASD groups (F = 13.92, p = .0001). There was no significant mean difference between the two ASD groups in NV cognitive age equivalency (p > .05).

Examining ADHD comorbidity by ascertainment group

To determine if proband/early intervention participants and Sibs-ASD were equally represented in the ASD+ADHD group, we conducted crosstab analyses. A total of 12 children (34%) from the combined proband/early intervention group and 6 children (22%) from the Sibs-ASD group were represented in the ASD+ADHD group. Chi-square analyses

revealed that this was not a significant difference ($\chi^2 = 1.077$, $p > .05$). Moreover, the two groups did not differ significantly in the percentage of children meeting clinical cutoffs on the Inattentive and Hyperactive subscales of the BASC-2, with 18.5% and 20% of each group meeting the cutoff on the Inattentive subscale ($\chi^2 = 0.021$, $p > .05$) and 14.8% and 22.9% meeting the cutoff on the Hyperactivity subscale, respectively ($\chi^2 = 0.632$, $p > .05$).

Between-group comparisons of internalizing psychopathology, social impairment and autistic mannerisms, and adaptive functioning

Next, we examined between-group differences in psychological functioning (using the BASC-2 Internalizing Composite Scale), social functioning (using the five SRS subscales), and adaptive functioning (using the three VABS-II subscales). One-way analysis of variance (ANOVA) was used in this analysis, with Bonferroni correction calculated for a total of nine analyses (0.05/9) resulting in $p \leq .005$ as the criterion for significance. Because ANOVA procedures assume equal variance between groups (Miller and Chapman, 2001: 45), we used the Levene Test of Homogeneity of Variances to test for unequal group variance in the dependent variables. The following dependent variables were found to have unequal group variance: the BASC-2 Internalizing Composite Scale, the SRS Social Awareness and Social Motivation subscales, and the VABS-II Daily Living subscale (*p*s < .05) (see Table 2).

Because there was unequal between-group variance on some of the dependent variables, we used the Welch Test of Equality of Means in lieu of the ANOVA F-statistic to test for significant between-group differences. With the exception of the BASC-2 Internalizing Composite Scale, all the between-group comparisons were statistically significant ($p < .001$). Next, Bonferroni post hoc tests were conducted to determine which groups accounted for the significant differences in the dependent variables. In Table 3, the means, standard deviations, Welch statistics, and degrees of freedom (*df*) for the dependent measures in these analyses are presented. Significant between-group differences using post hoc Bonferroni multiple comparisons ($p < .05$) are designated with superscripts. Group means at or above clinically significant thresholds of impairment and those meeting subclinical thresholds are also delineated.

Internalizing psychopathology

There were no significant between-group differences on the BASC-2 Internalizing Problems Composite Scale ($p > .05$), with all groups scoring within normal limits.

Social impairment and autism mannerisms

On the SRS, there were significant between-group differences on all subscales ($p < .001$). Post hoc comparisons revealed that the No ASD group scored significantly lower (healthier) than the two ASD groups on all five SRS sub-scales (Social Awareness, Social Cognition, Social Communication, Social Motivation, and Autistic Mannerisms; *p*s < .001). The ASD +ADHD group scored significantly higher (more abnormally) than the ASD Only group on all subscales of the SRS ($p < .001$). Mean T-scores on all SRS subscales were in the healthy range for the No ASD group, in the mild to moderately impaired range for the ASD Only group, and in the severely impaired range for the ASD+ADHD group, representing a

continuum of impairment associated with increasing breadth of developmental psychopathology.

Adaptive functioning

For the VABS-II, Welch analyses revealed a significant between-group difference in all domains (Communication, Socialization, and Daily Living Skills; *p*s < .001). Post hoc comparisons revealed that the No ASD group scored significantly higher (healthier) than the ASD Only and the ASD+ADHD groups in all domains (Communication, Socialization, and Daily Living (*p*s = .0001)). The ASD Only group scored significantly higher than the ASD +ADHD group on all three domains of the VABS-II (Communication ($p = .008$), Socialization ($p = .045$), and Daily Living Skills ($p = .03$)). Mean standard scores on all VABS-II subscales were in the healthy range for the No ASD group and in the mild to moderately impaired range for the ASD Only group. For the ASD+ADHD group, mean scores were in the moderately impaired range in the Communication domain and in the severely impaired range in the Socialization and Daily Living domains, representing a continuum of impairment across the three groups.

Cognitive functioning in the two ASD groups

Although no significant mean NV cognitive age equivalency scores were found between the two ASD groups of children, we wished to investigate whether greater NV cognitive impairment was more often associated with ADHD comorbidity with ASD than with noncomorbid ASD. This was accomplished by creating subgroups of children with ASD defined as high functioning (HFA) or low functioning (LFA). Children who completed the SB-5 were assigned to the HFA group if their nonverbal IQ (NVIQ) was ≥70 and to the LFA group if NVIQ was <70. All 10 children who received the MSEL were automatically assigned to the LFA group. A chi-square analysis revealed that the ASD+ADHD group comprised a significantly higher percentage of children classified as LFA than the ASD Only group (61% vs 25%; Fisher's exact test, $p = .01$), indicating that more children in the ASD+ADHD group were cognitively impaired compared with the ASD Only group.

Discussion

This study focused on young school-aged children with ASD recruited from a research sample rather than a clinical source. Based on parents' report using a standardized instrument, we found that 29% of our sample of children with ASD also exhibited clinically significant levels of ADHD. This rate of comorbid ADHD in an ASD sample was considerably lower than rates reported in clinical samples of children with ASD (Gadow et al., 2006; Goldstein and Schwebach, 2004; Lee and Ousley, 2006; Sinzig et al., 2009). However, the rate of comorbid ADHD in our research sample was consistent with that reported in the only published study of a community sample of children and adolescents with ASD (Leyfer et al., 2006), despite the wider age range and older mean age in the Leyfer et al. (2006) sample, which did not include younger siblings of children with autism. Thus, our study identified ADHD symptoms in a younger group of children from a sample with a narrower age range than has been previously reported in the published literature. This has implications for diagnosticians and interventionists. Our findings indicate that young

school-aged children with ASD should be assessed for ADHD. If clinically significant ADHD symptoms are identified, and social development does not appear to be responding to intervention, changes in the intervention program (e.g. intensity, strategies, goals) may be required.

As hypothesized, our comorbid ASD+ADHD group was significantly more impaired in cognitive, social, and adaptive functioning than our ASD Only group. Similar to other researchers' reports of significant NVIQ differences in ASD with and without comorbid ADHD (e.g. Sinzig et al., 2009), we found that a significantly higher percentage of children with comorbid ASD+ADHD were classified as having significant cognitive delays (low functioning autism) than children with ASD only (61% vs 25%). Parents also rated our participants with comorbid ASD+ADHD significantly higher in autism mannerisms (i.e. stereotypic and repetitive behaviors) than participants with ASD only. These findings are consistent with Yerys and colleagues' (2009) report of elevated autistic traits in children with ASD accompanied by comorbid ADHD. Our findings also are consistent with others' reports of more severe social problems (Holtmann et al., 2007) and maladaptive behaviors (Yerys et al., 2009) in children with ASD comorbid with ADHD than children with ASD only. These findings suggest that ADHD comorbidity may constitute a distinctive phenotype of ASD, as yet unidentified as such, and that these children may be at higher risk of social impairment and adjustment problems. There is some support for this hypothesis in the literature. For example, IQ has been found to be a significant moderator of attention problems and impulsivity in children with ADHD (Buchmann et al., 2011). Furthermore, Yerys and colleagues (2009) reported on elevated rates of externalizing problem behaviors as well as greater impairment in executive functioning in children comorbid with ASD +ADHD, compared with children with ASD only. Further research is needed to determine whether an ASD/ADHD phenotype, with its own distinguishing characteristics, may help explain some of the variance between children with ASD with and without ADHD on measures of psychopathology and social and adaptive functioning.

A strength of our study was the use of a more restricted age range of 4 through 8 years, serving to constrain age-related variation in ADHD symptomatology in our sample of children with ASD, thereby allowing for a more focused developmental analysis. Indeed, our findings of equal numbers of children meeting clinical cutoffs for the two subtypes of ADHD (inattention and hyperactivity) differ from previous reports in the literature. In previous studies, more children met criteria for the inattentive than the hyperactive subtype of ADHD (e.g. Lee and Ousley, 2006). The discrepancy between our findings and others' reports on this matter may be due to the younger, more confined age range of participants in the current study. Indeed, Lee and Ousley (2006) found a negative correlation between age and hyper-activity, and Gadow et al. (2006) reported that parents of younger children (ages 3–5 years) reported significantly higher rates of hyperactivity than did parents of older children (ages 6–12 years), supporting the expectation that a higher percentage of children in our restricted age range would meet clinical cutoffs for hyperactivity than studies where the age range extends into adolescence.

Contrary to expectations based on previous research (e.g. Gillott et al., 2001; Holtmann et al., 2007; Yerys et al., 2009), there were no group differences in parent ratings of symptoms

of mood and anxiety disorders in the present study; in fact, none of the groups demonstrated elevated rates of internalizing problems. One explanation for this finding may be that symptoms of inattention or hyperactivity may overshadow symptoms of anxiety in younger school-aged children, masking them from parents' observation. In addition, anxiety may be present but may be contextually specific at this young age. Such a possibility is raised by the report that teachers were more likely to rate children with ASD+ADHD as having symptoms of anxiety than were parents (Guttmann-Steinmetz et al., 2010).

Another issue that must be considered is that some previous studies (e.g. Holtmann et al., 2007) used different measures of internalizing problems, such as the Child Behavior Checklist (CBCL; Achenbach, 1991). However, studies testing the construct validity of the BASC-2 with the CBCL identified moderate to high correlations between the internalizing and externalizing problems behavior scales on the two instruments (i.e. .78 and .75, respectively; Reynolds and Kamphaus, 2004). Thus, an alternative explanation may be that our young sample had not aged into the manifestation of psychopathology (specifically those classified as internalizing problems). The median age of diagnosis of anxiety disorders in the general, nondisabled population is 11 years (Kessler et al., 2005), and rates of anxiety and depression in children with autism reportedly increase with age (e.g. Mayes et al., 2011). Thus, participants in the current study may later develop internalizing disorders (i.e. anxiety and depression). This could occur as part of an unfolding neurobiology or could involve an environmental component that initiates or exacerbates the onset of anxiety. For example, secondary to increased self-awareness of their deficits and with increasing age, encounters involving social challenges with peers may be experienced as more troubling (Chamberlain et al., 2007). In addition, internalizing problems may be difficult to discern in young children with ASD as they may not have conscious awareness of their internal emotional states, and even if they have some level of awareness, they may have difficulty conveying their emotional state to others due to their ASD-related communication impairments (Hobson and Meyer, 2005).

Limitations of the study

There were several limitations to the current study. First, we did not include a comparison group of non-ASD children with ADHD. The nine non-ASD children who met classification criteria for ADHD comprised a very small group that would not have allowed us to make meaningful group comparisons. Moreover, seven of these children were siblings of children with autism, and epidemiological and ASD family studies report an increased genetic liability for both ASD and ADHD in these children (Lichtenstein et al., 2010; Rommelse et al., 2010; Ronald et al., 2008) as well as an increased risk for social impairment (Constantino et al., 2010; Landa et al., 2007). Thus, these siblings may represent an intermediate phenotype involving social difficulties and mild psychopathology, masking between-group differences on the SRS and BASC-2. Future studies of ASD+ADHD comorbidity should include an ADHD group with no family history of ASD. This would permit a clearer understanding of how ADHD impacts the phenotype of ASD in comorbid cases without increased risk for additional impairments.

The second limitation was the categorization of participants into ADHD groupings based on parent report via a questionnaire rather than a clinical interview. However, in support of our findings, the measure we used to define the presence of ADHD (BASC-2) is based on *DSM-IV-TR* diagnostic criteria, and a similar rate of ADHD in a community ASD sample (31%) was identified in a previous study through direct clinical parent interview (Leyfer et al., 2006). A related issue is that *DSM-IV-TR* diagnostic criteria for ADHD require that symptoms be present in at least two different settings, and our measure was limited to one setting and one informant. Thus, future studies collecting data from other informants and other settings (such as teachers and schools) are needed to increase support for the findings of the current study. It is also important to obtain ratings from professionals who are knowledgeable in child development, as parents may tend to over- or under-rate hyperactivity and other attentional problem behaviors due to lack of understanding of the appropriateness of such behaviors at different ages and stages of development.

Conclusions and directions for future research

This study lends support to the growing consensus among autism researchers that a considerable proportion of children with ASD also meet diagnostic criteria for ADHD, and thus, these two disorders may indeed co-occur (e.g. Gadow et al., 2006; Holtmann et al., 2007). There are proposed changes to the fifth edition of the *Diagnostic and Statistical Manual of the American Psychiatric Association* (*DSM-V*) to remove the existing prohibition of a dual diagnosis of ASD and ADHD (www.dsm5.org). However, until the new edition of the *DSM* is published, it is imperative to begin to recognize the high co-occurrence rates of these two disorders as well as the potential increased risk for social and adaptive impairment associated with comorbidity of ASD and ADHD. Following the work of other researchers and building on the findings of the current study, more research in ASD +ADHD comorbidity is needed to further clarify the characteristics of the ASD+ADHD phenotype so that specialized treatments and interventions may be designed to improve outcomes and quality of life for this subgroup of children. This is important because children comorbid with the two disorders may have a higher risk for suboptimal outcomes and may benefit from different treatment methods or intensities than those with ASD only (e.g. Antshel et al., 2011). A major contribution to the field would be the prospective, longitudinal study of developmental psychopathology, from infancy, to understand the relative timing, stability, and nature of disruption in attention, social, communication, and cognitive processes in children at high risk of the ASD+ADHD phenotype and to possibly reveal protective factors.

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Table 1

Participant demographic information.

	No ASD/ADHD	ASD Only	ASD+ADHD
	n **= 91**	***n*** **= 44**	***n*** **= 18**
Mean age (in years)	5.54 (1.3)	5.59 (1.2)	5.67 (1.4)
Males*	43 (47%)^{a}	33 (75%)^{b}	18 (100%)^{c}
% Caucasian	79 (87%)	40 (91%)	14 (78%)
Hollingshead mean SES	57.05 (8.05)	57.34 (7.6)	56.22 (8.27)
Mean NV cognitive age equivalency* (in months)	75.20 (19.1)^{a}	59.20 (22.85)^{b}	50.78 (31.72)^{b}

ASD: autism spectrum disorder; ADHD: attention deficit hyperactivity disorder; SES: socioeconomic status; NV: nonverbal.

Hollingshead SES range: major business and professional (55–66) and medium business, minor professional, and technical (40–54).

^{a,b}Different superscripts designate significantly different between-group mean scores; groups with same superscripts are not significantly different from each other.

*$p < .001$.

Table 2

Test of homogeneity of variances on the dependent variables.

	Levene statistic	*p*	*df*
BASC-2			
Internalizing Composite	0.766	.467	2,148
Scale Score			
Social Responsiveness Scale			
Social Awareness	0.167	.846	2138
Social Cognition*	4.35	.015	2138
Social Communication*	9.58	.0001	2138
Social Motivation	0.184	.163	2138
Autistic Mannerisms*	3.34	.038	2138
Vineland Adaptive Behavior Scale			
Communication*	9.06	.0001	2146
Socialization*	7.42	.001	2146
Daily Living Skills	2.71	.07	2146

BASC-2: Behavioral Assessment System for Children–Second Edition.

*$p < .05$ = unequal variance in comparison groups.

Table 3

Between-group differences in parent and clinician ratings on the dependent variables.

	No ASD or ADHD	ASD Only	ASD+ADHD	Welch statistic
	n **= 91**	***n*** **= 44**	***n*** **= 18**	
BASC-2 Internalizing	*M* (SD)	*M* (SD)	*M* (SD)	(*df* = 2148)
Composite Scale score				
Composite Scale	48.1 (10.2)[a]	48.7 (11.3)[a]	49.9 (13.2)[a]	0.16
Social Responsiveness Scale				(*df* = 2138)
Social Awareness*	45.1 (10.2)[a]	68.1 (12.5)[b,d]	**76.8(10.5)**[c]	95.47
Social Cognition*	46.2 (9.3)[a]	69.0 (12.7)[b,d]	**77.0 (9.8)**[c]	102.95
Social Communication*	46.4 (9.3)[a]	69.4 (14.5)[b,d]	**78.6 (8.7)**[c]	116.05
Social Motivation*	48.3 (10.3)[a]	63.3 (12.7)[b,d]	**72.2 (12.9)**[c]	39.39
Autistic Mannerisms*	48.1 (10.4)[a]	69.3 (14.3)[b,d]	**83.5 (9.1)**[c]	115.05
Vineland Adaptive Behavior Scale				(*df* = 2146)
Communication*	109.3 (11.73)[a]	91.1 (17.8)[b]	74.0 (18.9)[c,d]	42.27
Socialization*	103.1 (10.5)[a]	80.3 (14.7)[b,d]	**69.3 (15.5)**[c]	69.71
Daily Living Skills*	98.9 (13.0)[a]	81.4 (14.5)[b,d]	**68.2 (17.3)**[c]	40.15

ASD: autism spectrum disorder; ADHD: attention deficit hyperactivity disorder; BASC-2: Behavioral Assessment System for Children–Second Edition; SRS: Social Responsiveness Scale; VABS-II: Vineland Adaptive Behavior Scales–Second edition.

Scores in the severely impaired range on the SRS and in the low range on VABS-II (≥2SD below the mean) are designated in bold.

[a,b,c] Different superscripts designate significantly different between-group mean scores using Bonferroni post hoc multiple comparisons ($p < .05$); groups with same superscripts are not significantly different from each other.

[d] Scores in the mild-to-moderately impaired range on the SRS and in the moderately low range (≥1 and <2SD below the mean) on the VABS-II.

* $p < .005$ (corrected p using Bonferroni correction).