Published in final edited form as:

Res Autism Spectr Disord. 2017 July ; 39: 20–32. doi:10.1016/j.rasd.2017.04.003.

Parent-Reported Problematic Sleep Behaviors in Children with Comorbid Autism Spectrum Disorder and Attention-Deficit/Hyperactivity Disorder

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Abstract

Background—Sleep problems are frequent and well documented in children with Autism Spectrum Disorders (ASD), children with Attention Deficit/Hyperactivity Disorder (ADHD) and children with internalizing problems, however limited work has examined sleep problems in children presenting with comorbid ASD/ADHD. In healthy children, sleep problems negatively impact social, emotional, and academic functioning. The current study sought to examine diagnostic severity as predictors of sleep problems in children with comorbid ASD/ADHD. Additionally, the association between sleep and “real-life” functional domains (i.e., intellectual functioning, academic achievement, and executive functioning) were assessed.

Method—Sleep, internalizing difficulties, intellectual functioning, academic achievement and executive functioning were assessed in 85 children with who carried the dual diagnoses of ASD and ADHD.

Results—Internalizing difficulties, rather than ASD or ADHD symptom severity, was the most consistent predictor of problematic sleep behaviors (i.e., nightmares overtiredness, sleeping less than other children, trouble sleeping, and Total Problematic Sleep Behaviors) in this sample. Further, parent report of problematic sleep behaviors was significantly associated with functional domains after controlling for ASD, ADHD, and internalizing symptoms.

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Conflict of Interest: We have no conflicts of interest to report with regard to this study. Dr. Pearson has received travel reimbursement and research support from the Forest Research Institute and Curemark LLC; Dr. Pearson has also served as a consultant to Curemark LLC and United BioSource Corporation (now Bracket). She has also received research support from Biomarin and Novartis.

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Conclusions—Results suggest that internalizing symptoms are associated with problematic sleep behaviors in children with comorbid ASD/ADHD and may have implications for the “real-life” functioning among children with comorbid ASD/ADHD.

Keywords

sleep; autism spectrum disorder; attention-deficit/hyperactivity disorder; child; psychopathology

Problematic Sleep Behaviors in Children with Comorbid Autism Spectrum Disorder And Attention-Deficit/Hyperactivity Disorder Adequate sleep is essential for healthy child development (Dahl, 1996a). By 2 years old, a child has spent approximately 14 months of life asleep (Anders, Sadeh, & Appareddy, 1995), their brain has reached 90% of its adult size (Chugani, Phelps, & Mazziotta, 1987), and several core developmental milestones have typically been achieved (e.g., walking, talking; Carruth, Ziegler, Gordon, & Hendricks, 2004; Raviv, Kessenich, & Morrison, 2004). The parallel between significant developmental changes and the need for sleep in the first two years of life suggests that sleep plays an integral role in human development (Dahl, 1996a). In fact, sleep problems in children are associated with impairments in academic or occupational achievement (Dewald, Meijer, Oort, Kerkhof, & Bögels, 2010), emotional and behavioral functioning (Pesonen et al., 2010), family cohesion (Bell & Belsky, 2008; Simard, Nielsen, Tremblay, Boivin, & Montplaisir, 2008), and neuropsychological abilities (Carskadon, Harvey, & Dement, 1981). As such, children with developmental and/or psychological disorders known to impact sleep, such as autism spectrum disorder (ASD), are at risk for further problems with “real-life” functioning (i.e., executive functioning, academic achievement, intellectual functioning) associated with sleep problems. Internalizing problems, defined here as inwardly focused negative thoughts or behaviors, such as fearfulness, social withdrawal and somatic complaints that are core components in the development of anxiety and/or depressive symptoms, are also highly associated with sleep problems (Alfano, Ginsburg, & Kingery, 2007; Liu et al., 2007). Further, internalizing problems have been shown to impact functional outcomes in healthy children (Cabeza & Nyberg, 2000; Wolfson & Carskadon, 2003).

Sleep in Children with Autism Spectrum Disorders

Children with ASD have difficulties with social interaction and communication, and demonstrate restricted and repetitive behaviors and interests (DSM-5; American Psychiatric Association, 2013). Due to the pervasive nature of ASD, families of children with ASD experience greater stress than parents of typically-developing children (Eisenhower, Baker, & Blacher, 2005). This increased stress, however, is magnified when a child with ASD experiences sleep problems (Lopez-Wagner, Hoffman, Sweeney, Hodge, & Gilliam, 2008). Sleep problems are one of the most prevalent difficulties affecting children with ASD (Ming, Gordon, Kang, & Wagner, 2008), occurring in up to 80% of children with the disorder (Liu, Hubbard, Fabes, & Adam, 2006; Wiggs & Stores, 2004b). Furthermore, sleep problems are consistently associated with greater ASD symptom severity (Cortesi, Giannotti, Ivanenko, & Johnson, 2010; Mayes & Calhoun, 2009). For example, behavioral and emotional difficulties common to children with ASD, including oppositional behavior, aggression, explosiveness,

attention deficit, impulsivity, hyperactivity, anxiety and depression, and mood variability are linked with greater sleep problems (Mayes & Calhoun, 2009). Prior research has suggested that the high prevalence of internalizing comorbidities among children with ASD further complicate observed sleep problems within this population (Cortesi, Giannotti, Ivanenko, & Johnson, 2010). Within specific internalizing symptoms domains, findings suggest there are links between sensory problems, sleep, and anxiety in children with ASD (Mazurek & Petroski, 2015), while research examining mood has revealed similar associations between insomnia and mood among young adults with high functioning ASD (Tani et al., 2003).

Internalizing Problems in Children with ASD

Children with ASD often present with comorbid internalizing problems (Simonoff et al., 2008; Joshi et al., 2010). Among children with ASD, current prevalence estimates of anxiety range from 11%-84% (White, Oswald, Ollendick, & Scahill, 2009), while rates of depressive disorders fall around 56% (Joshi et al. 2010). ASD severity and internalizing symptom severity are positively correlated (Mayes, Calhoun, Murray, & Zahid, 2011) and increased levels of internalizing psychopathology in young adults with ASD are associated with greater prevalence of sleep problems (Tani et al., 2003; Tani et al., 2004). It is unclear, however, to what extent internalizing psychopathology accounts for sleep disruptions observed in children with ASD. The interplay between oversensitivity to the environment due to sensory issues, emotional dysregulation and elevated internalizing symptoms may be further compounded by the inattention and hyper arousal symptoms of ADHD among children with comorbid ASD and ADHD.

ADHD in Children with ASD

In addition to more frequent sleep problems, up to 75% of individuals with ASD also exhibit ADHD symptoms (Frazier et al., 2001; Goldstein & Schwebach, 2004; Mayes, Calhoun, Mayes, & Molitoris, 2012; Pearson et al., 2006). Until recently, the dual-diagnoses of ASD and ADHD was not acknowledged by DSM diagnostic criteria, but changes in the DSM-5 (American Psychiatric Association, 2013) recognize an ASD/ADHD comorbid clinical presentation. Similar to children with ASD, sleep complaints are common among children with ADHD (Sung, Hiscock, Sciberras, & Efron, 2008). Notably, problems with sustained attention, distractibility, and impulsivity, core symptoms of ADHD (American Psychiatric Association, 2013) overlap with functional impairments observed in sleep deprived children (van der Heijden, Smits, & Gunning, 2005). Common pharmacological treatments for ADHD (e.g., psychostimulants) can give rise to or exacerbate sleep problems (van der Heijden et al., 2005) including greater difficulty falling asleep and greater tiredness upon waking (Kaplan, McNicol, Conte, & Moghadam, 1987). Although subjective parent reports vary, objective data (using polysomnography [PSG] and actigraphy) reveal increased motor movements as the only consistent difference between the sleep of children with ADHD compared to healthy controls (Corkum, Tannock, Moldofsky, Hogg-Johnson, & Humphries, 2001; Okada et al., 2013). These mixed findings may be due to bedtime problems, including bedtime resistance and “curtain calls” (i.e., calling the parent back for one more kiss/hug/goodnight/etc.; Corkum et al., 2001), as well as individual differences in physiological arousal (Okada et al., 2013).

Sleep in Children with Comorbid ASD and ADHD

Although approximately 74% of children with ADHD (Sung et al., 2008) and 80% children with ASD (Liu et al., 2006; Wiggs & Stores, 2004b) have been reported to experience sleep problems, there has been limited investigation of sleep in children with both diagnoses. Recent research examining sleep among children with comorbid ASD/ADHD has noted that sleep problems for children with comorbid ASD/ADHD does not differ from children with ADHD alone (Green, Sciberras, Anderson, Efron, & Rinehart, 2016; Thomas, Monahan, Lukowski, & Cauffman, 2015). Thomas and colleagues (2014) demonstrated that both internalizing and externalizing problems are a risk factor for elevated sleep problems for children with comorbid ASD/ADHD (Thomas et al., 2014), however this association was no longer significant after family SES variables (e.g., parent age, high school education) were taken into account. Furthermore, the association between sleep problems and internalizing and externalizing problems was similar relative to children with ADHD only (Thomas et al., 2014). Green and colleagues (2015) also examined parent report of ASD symptoms across children with and without a diagnosis of ADHD. Although higher ASD symptom severity was associated with overall poorer functioning (i.e., internalizing and externalizing problems, more peer problems and poorer quality of life) among children with ADHD, sleep problem severity was similar across both ADHD and non-ADHD groups (Green et al., 2016). Together, findings from preliminary research examining sleep in comorbid ASD/ADHD suggest that sleep problems within this comorbid population do not differ from sleep problems seen among children with ADHD only, however preliminary studies have yet to examine these links among a sample of carefully evaluated children with formally evaluated and diagnosed ASD and ADHD.

When sleep problems are compared between children with ASD and children with ADHD, children with ASD experience significantly more sleep problems than do children with either ADHD-Predominantly Inattentive presentation or ADHD-Combined presentation (Mayes, Calhoun, Bixler, & Vgontzas, 2009). Given the high occurrence of sleep problems in children in both ASD and ADHD clinical populations (Liu et al., 2006; Sung et al., 2008; Wiggs & Stores, 2004b), children having both ASD and ADHD may be at particularly high risk for sleep problems and related functional impairments (e.g., academics, executive functioning).

Internalizing Problems in Children with ADHD

Children with ADHD also experience elevated levels of internalizing problems with approximately 25% of these children meeting criteria for a co-occurring anxiety disorder and 15-75% meeting criteria for a co-occurring mood disorder (Biederman, Newcorn, & Sprich, 1991). Among children with ADHD, sleep problems are significantly related to internalizing psychopathology (Mayes, Calhoun, Bixler, Vgontzas, et al., 2009; Mick, Biederman, Jetton, & Faraone, 2000). After controlling for anxiety, previously observed associations between ADHD and sleep problems are no longer significant (Mick et al., 2000). Similarly, when Hansen and colleagues (2011) compared children with comorbid anxiety and ADHD to children with anxiety disorders only and children with ADHD only, the comorbid anxiety/ADHD group and the anxiety-only group experienced significantly

greater sleep problems than children with ADHD alone (Hansen, Skirbekk, Oerbeck, Richter, & Kristensen, 2011). Taken together, findings suggest that internalizing psychopathology plays an integral role in the presence of sleep problems across children with ADHD. However the contribution of internalizing problems to sleep problems among children with comorbid ASD and ADHD requires further investigation.

Internalizing Problems and Sleep

Internalizing psychopathology (i.e., anxiety, depression) has been closely linked with sleep disruption. In clinically anxious and depressed youth, rates of sleep disturbance have been estimated at 55% and 72.7%, respectively (Alfano, Ginsburg, & Kingery, 2007; Liu et al., 2007). Persistent sleep problems in childhood have been shown to predict adolescent and adult anxiety (Gregory & O'Connor, 2002), and acute sleep restriction has been linked with increased problems in emotional functioning (Dagys et al., 2012). While sleep problems are clearly linked with internalizing psychopathology, the vague nature of the term “sleep problems” typically used in the literature disallows for a more precise understanding of specific relationships worthy of potentially different intervention targets (i.e., trouble falling asleep may warrant sleep hygiene interventions, while frequent awakenings may warrant sleep disordered breathing assessment). Anxiety and depression are common problems in both children with ASD (Ghaziuddin, 2002; Joshi et al., 2010) and children with ADHD (Biederman et al., 1991; Smalley, et al., 2007), are closely tied to the arousal systems (Dieleman et al., 2015), and are likely related to sleep problems within these clinical populations.

“Real-Life” Functioning in Children with ASD and ADHD

Both children with ASD and ADHD struggle with executive functioning (Pennington & Ozonoff, 1996; Russell, 1997), but available research has not provided a clear distinction between executive functioning deficits across individuals with ASD and ADHD. In some studies, global executive functioning deficiencies have been observed for children with ASD (Geurts, Verté, Oosterlaan, Roeyers, & Sergeant, 2004), while other studies have observed only problems with planning and working memory among children with ASD (Happé, Booth, Charlton, & Hughes, 2006). For children with ADHD, some studies show better executive function relative to ASD groups, with only inhibition and verbal fluency problems noted for children with ADHD (Geurts et al., 2004). Conversely, other studies suggest children with ADHD struggle with flexibility related components of executive functioning over and above executive functioning difficulties observed in ASD (Happé et al., 2006). While there is no consensus on the severity of executive function problems observed across children with ASD and ADHD, available research has also not examined children who meet diagnostic criteria for both disorders. This omission may have reduced the variability of executive functioning skills via elimination of greater contrast in diagnostic clinical severity (Happé et al., 2006). The current study aims to build upon this limitation, by incorporating both ASD and ADHD diagnostic severity as predictors of “real-life” outcomes, including executive, intellectual and academic functional domains.

“Real-Life” Functioning and Sleep

Among healthy children, the association between sleep loss and neurological, academic, and behavioral functioning is well established. On the neurobiological level, sleep deprivation amplifies activity in reward emotion-linked networks of the brain (Yoo et al., 2007), leading to increased emotional lability (Gujar, Yoo, Hu, & Walker, 2011). Specifically, sleep loss has been linked with impaired communication between emotion areas and the prefrontal cortex (PFC; Yoo et al., 2007). As the PFC is well known to be associated with executive function (Cabeza & Nyberg, 2000), it is likely that executive function difficulties among children with comorbid ASD and ADHD would be further amplified by sleep related problems. Among healthy children, shorter sleep times on school nights is also associated with lower grades (Wolfson & Carskadon, 2003).

Children with ASD and ADHD often present with comorbid anxiety (Biederman, Newcorn, & Sprich, 1991; Simonoff et al., 2008; Smalley et al., 2007) and depression (Joshi et al., 2010; Smalley et al., 2007), and comorbid internalizing disorders are also highly prevalent in children with comorbid ASD and ADHD (Chen et al., 2015). Both partial sleep restriction and sleep deprivation in healthy adults is associated with difficulties in the emotional processing of facial expression and emotion related working memory skills (Chuah et al., 2010; Motomura et al., 2013). As children with ASD and ADHD already struggle with executive functioning and commonly experience comorbid internalizing disorders, sleep difficulties may further amplify the extent to which their executive functioning skills suffer on a day to day basis.

The Current Study

The current study examined sleep patterns in a sample of children with comorbid ASD and ADHD. As the broad concept of “sleep problems” has been defined in varying ways across the literature (e.g., nightmares, extended sleep onset latency, quality of sleep, etc.), the present study examined *problematic sleep behaviors* (i.e., observable behaviors relating to sleep or daytime sleepiness) in general, and on the item level. Examination of “sleep problems” with greater specificity allows for a clearer understanding of potential intervention areas. For example, nighttime fears can be effectively treated with a combination of CBT techniques (i.e., relaxation) and the introduction of a behavioral reward system, while stimulus control interventions are recommended for children with trouble falling sleep (Owens, France, & Wiggs, 1999). Our primary aim was to determine which diagnostic factors were related to problematic sleep behaviors in this sample. Within this aim, we hypothesized that increased ASD, ADHD, and internalizing symptom severity would predict elevated levels of problematic sleep behaviors. Our second aim was to assess the association between problematic sleep behaviors and functional impairment in children with ASD/ADHD specific to academic performance, intellectual functioning, and executive functioning. We expected greater severity of problematic sleep behaviors would predict increased academic, cognitive, and executive functioning difficulties after accounting for ASD and ADHD diagnostic severity and internalizing symptom severity.

Method

Participants

The sample included 85 children (M age = 9.29, SD = 1.79; range = 6.66 – 12.91; 64 boys) who met DSM-IV-TR criteria for both ASD and ADHD. Although DSM-IV-TR did not permit clinical diagnosis of both conditions in the same child, for the purpose of this research we identified those who met criteria for both. Tables 1 and 2 include detailed demographic and comorbidity information. Psychiatric diagnoses (including ADHD) were determined by the Diagnostic Interview for Children and Adolescents-IV (DICA-IV; Reich, 2000), and by a clinical interview with a licensed psychologist (DAP). A complete description of the larger protocol is published elsewhere (Pearson, et al., 2012, 2013). ASD diagnosis was confirmed using the Autism Diagnostic Interview, Revised (ADI-R; Rutter, Le Couteur, & Lord, 2003) and the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 1999). The ADI-R and ADOS were administered by personnel who were research-certified on these instruments. ADHD diagnoses were confirmed using both the DICA-IV and a t-score of 65+ on the ADHD Index of the both the Conners Parent and Teacher Rating Scales-Revised (Achenbach & Rescorla, 2001).

Measures

Demographics—Information collected from the parent detailing child age, sex, race, ethnicity, parent education, and socioeconomic status (via Hollingshead 4 factor SES; Hollingshead, 1975) was collected using questionnaires and during the clinical interview with the last author (DAP).

Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2001)—The CBCL is a parent-reported questionnaire that assesses a variety of behavioral and emotional problems encountered in school-age children. It has been shown to be effective for assessing behavioral problems in children with Autism (Noterdaeme, Amorosa, Mildenerger, Sitter, & Minow, 2001; Pearson et al., 2012). On the CBCL, parents were asked to rate the occurrence of seven sleep-related behaviors: nightmares, excessive overtiredness, sleeping more than most children, sleeping less than most children, trouble sleeping, bed wetting, and walking or talking during sleep. As parasomnias can be associated with medically-based issues (e.g., obstructive sleep apnea, rather than problematic sleep behaviors) and are considered developmentally appropriate in younger children, only the five non-parasomnia behavioral sleep items (i.e., nightmares, excessive overtiredness, sleeping more than most kids, sleeping less than most kids, and trouble sleeping) on the CBCL were included in the present study. Parents rated each individual item on a 3-point scale (0=not true; 1=sometimes true; 2=always true). Raw frequencies and mean scores on the 3-point scale for endorsement of each item are presented in Table 3. Using similar procedures to Alfano, Ginsburg and Kingery (2007), the five sleep items were re-coded as present (i.e., sometimes/always true = 1) or absent (i.e., not true = 0) and a Total Problematic Sleep Behavior Score was calculated by summing the five individual sleep items. Thus, this converted summary score could range from 0-5 (relative to the raw score range of 0-10). CBCL sleep variables have been found to correspond with objective sleep measures in previous research (Gregory et al., 2011).

The CBCL also produces several syndrome scales. To capture internalizing difficulties, the current study utilized the Internalizing Problems scale, which includes withdrawn/depressed, anxious/depressed and somatic problems syndrome scales (t-scores).

Autism Diagnostic Interview-Revised (ADI-R; Rutter et al., 2003)—The ADI-R is a clinician administered semi-structured interview with a parent or caregiver that assessed ASD symptomology. A series of algorithms produce final scores on scales of Reciprocal Social Interaction, Communication (verbal or nonverbal), Restricted Repetitive and Stereotyped Behaviors, and Abnormal Development. Similar to Uddin and colleagues (2011), the total raw score summed across these scales was used to measure ASD symptoms severity in the current study. Both the ADI-R individual subscale scores and the total raw score were utilized as measures of ASD severity.

Conners Parent Rating Scale-Revised (CPRS-R; Conners et al., 1997)—The CPRS-R is a widely used questionnaire that assesses ADHD symptomatology and behaviors closely associated with ADHD, including externalizing and internalizing symptoms. The Conners long form was used in the present study (CPRS-R:L, 80 items). The CPRS-R:L yields 14 subscales including oppositional, cognitive problems/inattention, hyperactivity, anxious-shy, perfectionism, social problems, psychosomatic, Conners Global Index (CGI): Total, CGI: Restless-Impulsive, CGI-Emotional Lability, and the ADHD index. The CPRS-R:L is normed for children 3-17 years of age (Conners et al., 1997), and yields both raw and t-scores, with higher scores being indicative of greater symptom severity. The CPRS-P ADHD Index t-score was used as a global measure of parent reported ADHD symptom severity, and the two subscales of Cognitive Problems/Inattention and Hyperactivity were used to assess the severity of the two ADHD clusters of “Inattentive” and “Hyperactive-Impulsive” behaviors.

Stanford-Binet Intelligence Scale, Fifth Edition (Roid, 2003)—The Stanford-Binet 5th Ed. is a widely used measure of intellectual ability that assesses Full Scale IQ (FSIQ), verbal IQ (VIQ), nonverbal IQ (NIQ). VIQ and NIQ scaled scores were used in the current study as intellectual functioning measures.

Wide Range Achievement Test (WRAT-4; Wilkinson & Robertson, 2006)—The WRAT-4 is a screening measure of academic achievement for individuals from 5 to 94 years of age (Wilkinson & Robertson, 2006). The WRAT-4 consists of four subtests: Word Reading, Sentence Comprehension, Spelling, and Math Computation. Standard scores from all four subtests were used in the current analyses as functional academic measures.

Behavioral Rating Inventory of Executive Functioning (BRIEF; Gioia, Isquith, Retzlaff, & Espy, 2002)—The BRIEF is an 86-item parent reported questionnaire designed to assess broad executive functioning in children between 5-18 years of age. The BRIEF comprises eight subscales: inhibit, shift, emotional control, initiate, working memory, plan/organize, organize materials, and monitor. These subscales form two broader scales of the Behavioral Regulation Index (BRI; includes inhibit, shift and emotional control subscales) and the Metacognition Index (MI; includes initiate, working memory, plan/

organize, organization of materials and monitor subscales). BRI and MI t-scores were used to assess executive function in this study.

Procedure

Archival data from a larger project examining response to stimulant treatment of ADHD in children with comorbid ASD and ADHD was utilized in the current study. Each child's primary caregiver completed the CPRS-R at home as part of the screening procedure. Participants with elevated scores were invited for an initial assessment to confirm comorbid ASD diagnosis and the presence of ADHD symptoms. ASD diagnoses were verified using the ADOS during the initial evaluation appointment. A subsample later completed a double blind medication trial (Pearson et al., 2013), however data used in the present study was collected during the pre-medication trial evaluation that assessed the child's clinical status. Thus, all measures of sleep, behavior, internalizing symptoms, ASD and ADHD were assessed prior to psychopharmacological treatment.

Statistical Approach

To address Aim 1, hierarchical linear regression analyses were completed to determine the relationship between internalizing symptom severity (as assessed by the Internalizing Problems subscale of the CBCL), ASD symptom severity (as assessed by the ADI-R total score), ADHD severity (as assessed by the t-scores on the Conners Cognitive/Inattention and Hyperactivity subscales of the ADHD Index), and Total Problematic Sleep Behavior (i.e., specific sleep items and Total Problematic Sleep Behavior score). A second set of hierarchical linear regressions were completed utilizing individual ADI-R subscales to examine the unique association between these subscales and each problematic sleep behavior. This approach allowed us to assess the relative contributions of each clinical predictor (i.e., internalizing psychopathology, ASD, ADHD symptoms) to the overall variance in problematic sleep behaviors. Covariates were entered into step 1, Internalizing symptoms were entered into step 2, ASD severity was entered into step 3, and ADHD severity was entered into step 4 in each model. To address Aim 2, multivariate regressions were used to examine the relationship between problematic sleep behaviors (i.e., the Total Problematic Sleep Behaviors Score) and functional domains (i.e., academic performance, intellectual functioning, executive functioning), using internalizing symptom severity (CBCL), ASD symptom severity (ADI-R total raw score) and ADHD symptom severity (Conners) as predictor variables. A second multivariate regression was completed utilizing individual ADI-R subscales to examine the unique relationship between these subscales, problematic sleep behaviors and functional domains. Parent report was used to assess sleep behaviors, ASD, and ADHD. Parent report was used as parents are most able to report on sleep and because concordance rates between parent and teacher ratings of ASD and ADHD are high (Pearson et al., 2012). This approach allowed us to assess the impact of sleep behavior on collective functional domains, rather than examining each of these constructs separately. Preliminary correlational analyses were also conducted to identify relevant covariates for subsequent analyses.

Power

Two power analyses were conducted using G*Power software (Faul, Erdfelder, Lang, & Buchner, 2007) for hierarchical linear regression. For Aim 1, an *a priori* power analysis with adequate power (.80; Cohen, 1988) and 5 predictors needed a sample size of $N = 45$. For Aim 2, an *a priori* power analysis with adequate power (.80; Cohen, 1988) and 3 predictors needed a sample size of $N = 38$. As such, the present study ($N = 85$) had adequate power to detect effects using the data analytic plan described above.

Results

Preliminary analyses revealed that father's educational level was significantly correlated with WRAT-4 subscales of word reading ($r = .38, p < .001$), sentence comprehension ($r = .27, p = .013$), and spelling ($r = .35, p = .001$). Thus, paternal education was entered as a covariate in the model assessing functional domains (Aim 2). Similarly, parent-reported child internalizing symptoms was significantly correlated with nonverbal IQ ($r = .23, p = .029$), academic measures of sentence comprehension ($r = .216, p = .047$), ADHD Symptom severity ($r = .232, p = .032$), and both measures of executive functioning (BRI: $r = .397, p < .001$; MI: $r = .313, p = .004$). Parent-reported externalizing symptoms (CBCL) was significantly correlated with nightmares ($r = .336, p = .002$), overtiredness ($r = .335, p = .002$), sleeping more than other kids ($r = .343, p = .001$), the total Sleep Problems Score ($r = .446, p < .0001$), WRAT-4 Sentence comprehension ($r = .233, p = .032$), ADHD symptom severity ($r = .395, p < .001$), and both measures of executive functioning (BRI: $r = .459, p < .001$; MI: $r = .446, p < .001$). Despite high correlations with predictors and outcome variables, CBCL externalizing problems were not included as a covariate due to conceptual overlap with study outcomes (i.e., ADHD symptom severity) and our conceptual focus on internalizing problems.

The frequency of problematic sleep behaviors in this sample is shown in Table 3. Children in the current sample had a broad range of sleep presentations, with 24.7% of the sample endorsing always sleeping more than other children, and 22.4% of the sample endorsing consistent trouble sleeping.

Predicting Problematic Sleep Behaviors from Severity of ASD and ADHD Symptoms

To address our first aim, we examined if ASD, ADHD, and internalizing symptom severity, as indexed by the ADI-R total raw score, the Conners ADHD Index, and the internalizing problems CBCL syndrome scale respectively, would make unique contributions to individual CBCL sleep behaviors. To test this aim, six hierarchical regression models were used to predict individual CBCL sleep behaviors with the CBCL internalizing problems subscale entered first (step 2), followed by ADI-R total raw score (step 3), and Conners ADHD (step 4). All six models were then run again utilizing ADI-R subscale scores (step 3) in place of the total ADI-R raw score.

In models utilizing ADI-R total raw scores, parent-reported child internalizing symptoms were significantly associated with parent-report of nightmares ($R^2_{change} = 0.109, F_{change} = 10.034, \beta = 0.330, p = .002$), being overtired ($R^2_{change} = 0.186, F_{change} = 18.487, \beta =$

0.431, $p < .001$), sleeping less than other children ($R^2_{change} = 0.069$, $F_{change} = 6.099$, $\beta = 0.263$, $p = .016$), trouble sleeping ($R^2_{change} = 0.059$, $F_{change} = 5.162$, $\beta = 0.242$, $p = .026$) and Total Problematic Sleep Behaviors score ($R^2_{change} = 0.218$, $F_{change} = 22.595$, $\beta = 0.467$, $p < .001$). After accounting for internalizing problems, none of the specific sleep behaviors nor the total sleep behavior score were significantly associated with ASD total severity score. After accounting for internalizing problems and ASD severity, the addition of ADHD severity to the model was not significantly associated with any specific sleep behavior, however there was a trend toward significance between cognitive/inattention problems and overtiredness ($\beta = 0.202$, $p = .074$). See Table 4 for hierarchical regression coefficients.

In models utilizing ADI-R subscales scores as a multivariate measure of ASD symptom severity, all parent reported child internalizing results remained consistent with findings reported above. After accounting for internalizing problems, ASD severity approached significance in association with being overtired ($R^2_{change} = 0.088$, $F_{change} = 2.320$, $F_{change}p = .064$); specifically, the Restricted-Stereotypes Behavior Total was associated with being overtired ($\beta = 0.43$, $p = .018$). Additionally, ASD symptom severity was significantly associated with sleeping more than other children ($R^2_{change} = 0.128$, $F_{change} = 2.908$, $F_{change}p = .027$); specifically, the Abnormality of Development Total was associated with lower scores on the sleeping more than other children item ($\beta = -.068$, $p = .008$). There was also a trend toward significance between Restricted-Stereotyped Behaviors total and nightmares ($\beta = -.042$, $p = .051$). After accounting for internalizing problems and ASD severity, the addition of ADHD severity to the model was not significantly associated with any specific sleep behavior, nor was there a trend between cognitive/inattention problems and overtiredness (as reported above).

Sleep Behaviors as Predictors of Functional Impairment

Our second aim was to examine the relation between problematic sleep behaviors on intellectual, academic, and executive functional domains, over and above internalizing symptoms, ASD and ADHD diagnostic severity (see Table 5 for correlations). To test this aim, a multivariate regression model was used to examine the relation between Total Problematic Sleep Behavior score on collective functional domains (i.e., intellectual, academic, and executive functioning). After controlling for paternal education, internalizing symptoms, ASD and ADHD severity the Total Problematic Sleep Behavior score ($Wilks\ Lambda = .787$, $F = 2.130$, $p = .045$, $Partial\ eta^2 = 0.213$) was significantly associated with multivariate functional domains (i.e., intellectual, academic, and executive functioning). Follow up univariate regression models were examined (See Table 6). Total Problematic Sleep Behavior was significantly associated with better sentence comprehension ($F = 6.029$, $p = .017$, $Partial\ eta^2 = .079$).

When this model was run with ADI-R subscales rather than ADI-R total raw score, the association between the Total Problematic Sleep Behavior Score and functional domains (i.e., intellectual, academic, and executive functioning) only approached significance ($Wilks\ Lambda = .782$, $F = 2.089$, $p = .051$, $Partial\ eta^2 = 0.218$), however the univariate association between Total Problematic Sleep Behavior and better sentence comprehension ($F = 6.23$, $p = .015$, $Partial\ eta^2 = .085$) remained significant.

Discussion

The present study examined the relation between ASD and ADHD symptom severity and specific parent-reported problematic sleep behaviors in children with comorbid ASD and ADHD diagnoses. Based on high rates of comorbidity and previous research linking internalizing psychopathology with sleep problems in children with ADHD and ASD (Chen, et al., 2015), we also examined the specific contribution of internalizing symptoms to problematic sleep behaviors in this sample of children with comorbid ASD and ADHD. The impact of problematic sleep behaviors on intellectual, academic, and executive functioning was also assessed. Our findings provide preliminary insight into the constellation of problematic sleep behaviors experienced by children with comorbid ASD and ADHD and the impact of these behaviors on intellectual, academic, and executive functioning.

Diagnostic Severity Predicting Problematic Sleep Behaviors

When examining diagnostic severity as a predictor of sleep behaviors, internalizing symptoms--rather than ADHD or ASD symptom severity--emerged as the most consistent predictor of individual and Total Problematic Sleep Behaviors. Specifically, the presence of internalizing symptoms in our comorbid sample was significantly associated with more nightmares, excessive sleepiness (i.e. being overtired), sleeping less than other children, trouble sleeping, and higher Total Problematic Sleep Behaviors. Although there was preliminary evidence linking ADHD symptoms with being overtired and linking higher repetitive/stereotypical ASD behaviors with being overtired, internalizing symptoms remained a significant predictor across all models. These results are consistent with prior research demonstrating high levels of comorbid internalizing problems in children with ASD (Simonoff et al., 2008), ADHD (Smalley, et al., 2007) and comorbid ASD/ADHD (Chen et al., 2015). Additionally, findings within the current study are similar to other recent studies demonstrating greater risk of sleep problems among children with comorbid ASD/ADHD in the presence of both internalizing and externalizing problems (Thomas, et al., 2015).

Internalizing psychopathology is associated with a variety of sleep problems (Gregory, Rijdsdijk, Dahl, McGuffin, & Eley, 2006). In children with ASD, those classified as “poor sleepers” demonstrate greater affective problems relative to healthy controls (Malow et al., 2006). Additionally, anxiety has been identified as the strongest predictor of sleep problems in ASD (Hollway, Aman, & Butter, 2013) and several studies have identified sleep problems in children with ASD as related to increased anxiety, increased reactivity to sensory stimulation (Malow & McGrew, 2008; Mazurek & Petroski, 2015), and hyper-arousal (Hollway & Aman, 2011; Richdale & Baglin, 2013). Consistent with this body of research, findings from the present study emphasize the role of internalizing problems in problematic sleep behaviors for children with ASD, and suggest that internalizing symptoms have a greater influence on nightmares and being overtired than do ASD and ADHD symptom severity alone. Although it was beyond the scope of this current study to objectively measure arousal, the underlying hyper-arousal seen in youth with ASD (Hollway & Aman, 2011; Richdale & Baglin, 2013) may partially explain the strong link between internalizing symptoms and sleep related problems seen in this sample. This relationship clearly warrants further investigation.

Although internalizing symptoms appeared to be the best predictor of increased overtiredness in the present study, inattention symptom severity did account for a portion of the variance in parental report of overtiredness. Notably, symptoms of hyperactivity were not associated with increases in overtiredness. These results suggest that, in addition to internalizing symptoms, inattention may be another factor associated with being overtired in children with comorbid ASD and ADHD. It is possible that inattention, rather than hyperactivity, is related to overtiredness, because more cognitive energy and focus may be needed to concentrate when symptoms of inattention are present. In sum, our results indicate that internalizing symptoms are the most consistent predictor of problematic sleep behavior in our comorbid ASD/ADHD sample, yet ADHD problems of inattention may also contribute to overtiredness.

Functional Impairment Findings

Only the Total Problematic Sleep Behavior score was significantly associated with intellectual, academic, and executive functioning domains in the present study. When each functional domain was examined individually, however, only a significant association between the Total Problematic Sleep Behavior score and improved sentence comprehension was observed. This finding was surprising as it is inconsistent with the broader sleep literature linking sleep problems in children with temporary impairments in academic achievement (Dewald, Meijer, Oort, Kerkhof, & Bögels, 2010) and neuropsychological abilities (Carskadon, Harvey, & Dement, 1981). Thus additional research is needed to further understand the association between sleep and academic achievement in children with comorbid ASD and ADHD. Overall, findings from the current study call for additional research examining the directionality of these links between sleep duration and the “real world” functional abilities in children with comorbid ASD and ADHD with greater emphasis on objectively assessed executive functioning skills.

Limitations

Despite the novel findings of the present study, there were limitations. Our relatively small sample was previously collected as part of a larger study, and therefore the measure of problematic sleep behaviors used in the current study was not purposefully collected. Our approach, however, is consistent with Alfano, Ginsburg and Kingery (2007), in which single CBCL items were used to assess sleep constructs. Additionally, problematic sleep behaviors were brief, only reported on by parents, and no measure of sleep duration was included. Future research should expand upon this approach by utilizing child-report of sleep (i.e., perceived sleep onset latency, sleep duration and sleep quality) and objective physiological measurements of sleep (i.e., actigraphy) to further examine the potential protective impact of sleeping “more than other children” in children with comorbid ASD and ADHD. In addition, the inclusion of a control group in future research will add to the understanding of how these relationships differ for youth with and without ASD/ADHD diagnoses. Future studies should also include specific sleep measures validated for children with ASD and ADHD. Our measure of anxiety (CBCL) only provided a broad view of anxiety/internalizing symptoms, and was not indicative of clinically significant anxiety problems. Although prior studies examining sleep in children with comorbid ASD/ADHD have utilized the combination of internalizing and externalizing problems (Thomas, et al., 2015; Green et al., 2016), inclusion

of broad externalizing problems was beyond the scope of the current study. Building on this work, continued research is needed to further examine the relative contribution of broad internalizing *and* externalizing problems within children who meet diagnostic criteria for both ASD and ADHD.

All measures were assessed at a single time point, therefore temporal precedence cannot be established. Future research should work toward a developmental and longitudinal approach to clarify the directionality of our results, as the current study cannot determine if poor sleep leads to worse academic, intellectual, and executive functioning for these children, or if poor functioning across these domains and problematic sleep behaviors are both products of increased ASD, ADHD, and anxiety severity. Lastly, although findings of the current study are consistent with previous research linking internalizing symptoms and sleep problems (Alfano, Beidel, Turner, & Lewin, 2006; Alfano et al., 2007; Alfano et al., 2010; Gregory et al., 2005; Gregory & Eley, 2005; Gregory et al., 2004; Gregory & O'Connor, 2002), the lack of findings across the other sleep items (e.g., trouble sleeping; sleeping less than other kids) may indicate that our sample was underpowered to detect small effect sizes. It will be important to recruit larger samples of children in future study to more fully explore these questions.

Conclusions & Implications

Our current findings suggest that internalizing symptoms play a significant role in problematic sleep behavior in children with ASD and ADHD. As sleep problems in this comorbid sample have been linked with greater behavioral symptoms and problems with family functioning (Lopez-Wagner et al., 2008; Mayes & Calhoun, 2009), the impact of internalizing psychopathology on sleep in ASD requires future study. Clinically, results from the current study imply children with ASD and ADHD who present with problematic sleep behaviors should receive a comprehensive evaluation to identify specific problematic sleep behaviors and their potential relationship with internalizing psychopathology as potential intervention targets. An individualized and targeted sleep or internalizing symptom intervention could then be developed to not only improve the child's sleep difficulties, but also enhance their broader functional abilities.

Acknowledgments

This research was supported by grant NIMH grant MH 072263, as well as Project T73MC22236 from the Maternal and Child Health Bureau. The authors are grateful to the families for their participation in this research. The authors also express their appreciation to Rosleen Mansor, M.A. for her support of this project.

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Highlights

- Internalizing symptom severity was most consistently associated with problematic sleep behaviors.
- ASD and ADHD symptom severity was not associated with problematic sleep behaviors.

Table 1
Demographic Breakdown of Current Sample

Variable	(n/%)
Female	21 (21.2)
Race/Ethnicity	
White	60(70.6)
Black	14(16.5)
Asian	3(3.5)
Other/Mixed Race	8(9.5)
Parental Marital Status	
Single	5(5.9)
Married	66(77.6)
Separated	3(3.5)
Divorced	10(11.8)
Co-morbid Psychiatric Diagnoses (DSM-IV-TR)	
ADHD-Combined Type	63 (74.1)
ADHD-Predominantly Inattentive Type	21 (24.7)
ADHD-Predominantly HA-Impulsive Type	1 (1.2)
Oppositional Defiant Disorder	18(21.2)
Conduct Disorder	2(2.4)
Major Depressive Disorder	1(1.2)
Dysthymic Disorder	2(2.4)
Separation Anxiety Disorder	1(1.2)
Enuresis	10(11.8)
Encopresis	4(4.7)
Generalized Anxiety Disorder	1(1.2)
Specific Phobia	28(32.9)
Social Phobia	5(5.9)
Obsessive Compulsive Disorder	12 (14.1)

Note. DSM-IV-TR diagnoses were determined using the Diagnostic Interview for Children and Adolescence-IV (DICA-IV; Reich, 2000)

Table 2
Demographic and Academic, Intellectual and Executive Functioning Measure Descriptive Statistics

Variable	Mean	Std. Deviation
Age in years (6.66 – 12.91yrs)	9.294	1.791
Mother's Educational Level (yrs.)	15.721	2.260
Father's Educational Level (yrs.)	15.969	3.012
Hollingshead 4-factor SES	49.476	13.037
Number of Comorbid Diagnoses	0.988	1.052
Total Medications	0.965	1.063
CBCL anxious/depressed t-score	61.400	8.919
ADI-R Sum of Subscales	48.082	11.075
CPRS ADHD index t-score	74.223	8.661
CPRS cognitive problems/inattention t-score	71.976	9.797
CPRS hyperactivity t-score	64.372	12.133
SB5 Nonverbal IQ	85.694	18.619
SB5 Verbal IQ	80.705	21.883
WRAT-4 Word Reading SS	89.847	18.500
WRAT-4 Sentence Comprehension SS	85.786	17.094
WRAT-4 Spelling SS	89.811	19.357
WRAT-4 Math Computation SS	81.352	17.522
BRIEF - BRI T Score - Parent	68.736	11.273
BRIEF - MI T Score - Parent	69.741	9.243

Note. CBCL (Child Behavior Checklist); ADI-R (Autism Diagnostic Interview-Revised); SB5 (Stanford-Binet Intelligence Scales - 5th Edition); SS (Standard Score); IQ (Intellectual Quotient); WRAT-4 (Wide Range Achievement Test – 4th Edition); CPRS (Connors Parent Rating Scale-Revised); SES (Socioeconomic status)

Table 3
Raw Frequency of Problematic Sleep Behaviors Items on the CBCL

Sleep Items	Not True n (%)	Sometimes True n (%)	Always True n (%)	Mean	SD
Nightmares	60 (70.6)	21 (24.7)	4 (4.7)	0.341	0.568
Overtired	67 (78.8)	15 (17.6)	2 (2.4)	0.226	0.474
Sleeping Less than Other Kids	43 (50.6)	21 (24.7)	21 (24.7)	0.741	0.833
Sleeping More than Other Kids	75 (88.2)	5(5.9)	5 (5.9)	0.177	0.515
Trouble Sleeping	44 (51.8)	22 (25.9)	19 (22.4)	0.706	0.814
Raw Total Sleep Behaviors				2.024	1.962

Note. CBCL (Childhood Behavior Checklist)

Table 4
Hierarchical Linear Regression Results for Sleep Items on the CBCL

Model – Dependent Variable	Variable	R ² Change	F Change	p	β	p
1 – Nightmares	Internalizing Problems	.109	10.034	.002*	.321	.004*
	ASD (ADI-R) Raw Total			.497	-.068	.521
	ADHD (CPRS)	.013	.594	.555		
2 – Overtired	Cognitive/Inattention				.009	.942
	Hyperactivity				.111	.340
	Internalizing Problems	.186	18.487	.000**	.384	.000**
	ASD (ADI-R) Raw Total	.011	1.123	.293	.115	.252
	ADHD (CPRS)	.032	1.637	.201		
3 – Sleeping Less than other Kids	Cognitive/Inattention				.202	.074 [†]
	Hyperactivity				-.078	.476
	Internalizing Problems	.069	6.099	.016*	.265	.018*
	ASD (ADI-R) Raw Total	.020	1.821	.181	-.142	.190
	ADHD (CPRS)	.011	.473	.625		
	Cognitive/Inattention				-.017	.888
4 – Sleeping More than other Kids	Hyperactivity				.110	.353
	Internalizing Problems	.012	1.031	.313	.062	.581
	ASD (ADI-R) Raw Total	.016	1.352	.248	.138	.210
	ADHD (CPRS)	.033	1.401	.252		
	Cognitive/Inattention				.139	.264
5 – Trouble Sleeping	Hyperactivity				.080	.507
	Internalizing Problems	.059	5.162	.026*	.234	.037*
	ASD (ADI-R) Raw Total	.014	1.273	.262	-.117	.281
	ADHD (CPRS)	.004	.161	.851		
	Cognitive/Inattention				.058	.633
6 – Total Problematic Sleep Behav.	Hyperactivity				.010	.936
	Internalizing Problems	.218	22.595	.000**	.435	.000**
	ASD (ADI-R) Raw Total	.004	.462	.499	-.061	.535

Model – Dependent Variable	Variable	R ² Change	F Change	p	β	p
	ADHD (CPRS)	.022	1.148	.323		
	Cognitive/Inattention				.108	.332
	Hyperactivity				.073	.499

Note: β corresponds to standardized β in a model with all variables included; CBCL (Child Behavior Checklist); ADHD (Attention-Deficit/Hyperactivity Disorder); ASD (Autism Spectrum Disorder); ADI-R (Autism Diagnostic Interview-Revised); CPRS (Connors Parent Rating Scale-Revised);

[†] < .10;
* < .05;
** < .01

Table 5
Bivariate Correlations between Sleep and Functional Domains

Correlations	Nightmares	Overtired	Sleep Less than Other Kids	Sleep More than Other Kids	Trouble Sleeping	Total Problematic Sleep Behav. Total Score
CBCL internalizing t-score	0.330 **	0.431 **	0.263 *	0.111	0.242 *	0.467 **
SB5 Nonverbal IQ	0.228 *	0.038	0.143 *	0.057	0.125	0.208
SB5 Verbal IQ	0.188	0.028	0.058	0.205	0.063	0.168
WRAT-4 Word Reading Standard Score	0.071	-0.086	0.137	0.086	0.068	0.097
WRAT-4 Sentence						
Comprehension Standard Score	0.193	0.200	0.222 *	0.185	0.138	0.311 **
WRAT-4 Spelling Standard Score	0.066	-0.005	0.093	0.145	0.034	0.102
WRAT-4 Math Computation Standard Score	0.154	-0.004	0.137	0.200	0.082	0.179
ADI-R Sum of Subscales	-0.050	0.130	-0.126	0.134	-0.106	-0.041
ADI-R Social Total	-.100	.094	-.160	.157	-.086	-.069
ADI-R Communication Verbal Total	.057	.067	-.040	.106	-.064	.019
ADI-R Communication Nonverbal Total	.057	.067	-.040	.106	-.064	.019
ADI-R Restricted-	.057	.067	-.040	.106	-.064	.019
Stereotyped Behaviors Total	-.131	.339 **	-.079	.188	-.122	.026
ADI-R Abnormality of Development Total	.034	-.133	-.085	-.232 *	-.079	-.153
CPRS ADHD index t-score	0.179	0.212	0.064	0.147	0.117	0.225 *
CPRS cognitive problems/inattention t-score	0.133	0.261 *	0.097	0.180	0.121	0.247 *
CPRS hyperactivity t-score	0.150	0.050	0.133	0.143	0.059	0.170
BRIEF - BRI T Score -Parent	0.174	0.319 **	0.187	0.139	0.092	0.295 **
BRIEF - MI T Score -Parent	0.244 *	0.256 *	0.079	0.064	0.082	0.235 *
Sleep Interrelations						
Nightmares	-					
Overtired	0.071	-				
Sleep Less than Other Kids	0.302 **	0.155	-			
Sleep More than Other Kids	0.002	0.362 **	-0.138	-		
Trouble Sleeping	0.146	0.215	0.732 **	-0.065	-	

Correlations	Nightmares	Overtired	Sleep Less than Other Kids	Sleep More than Other Kids	Trouble Sleeping	Total Problematic Sleep Behav. Total Score
Total Problematic Sleep Behav. Total Score	.0548**	0.554**	0.782**	0.277*	0.764**	-

Note. CBCL (Child Behavior Checklist); SB5 (Stanford-Binet Intelligence Scales - 5th Edition); SS (Standard Score); IQ (Intellectual Quotient); WRAT-4 (Wide Range Achievement Test – 4th Edition); CPRS (Connors Parent Rating Scale-Revised);

* < .05;
** < .01

Table 6
Univariate Follow-Up tests of 'Total Problematic Sleep Behaviors Score' on Functional Academic, Intellectual, and Executive Functioning Domains

Independent Variable	Dependent Variable	Type III Sum of Squares	df	Mean Square	F	Sig.	Partial Eta ²
Total Problematic Sleep Behaviors	SB5 Nonverbal IQ	844.019	1	844.019	2.728	.103	.038
	SB5 Verbal IQ	460.082	1	460.082	1.041	.311	.015
	WRAT-4 Word Reading SS	134.977	1	134.977	.475	.493	.007
	WRAT-4 Sentence Comprehension SS	1446.287	1	1446.287	6.029	.017*	.079
	WRAT-4 Spelling SS	204.914	1	204.914	.647	.424	.009
	WRAT-4 Math Computation SS	495.999	1	495.999	1.828	.181	.025
	BRIEF - BRI T Score - Parent	36.537	1	36.537	.399	.530	.006
	BRIEF - MIT Score - Parent	.736	1	.736	.017	.897	.000

Note. CBCL (Child Behavior Checklist); SB5 (Stanford-Binet Intelligence Scales - 5th Edition); SS (Standard Score); IQ (Intellectual Quotient); WRAT-4 (Wide Range Achievement Test - 4th Edition); CPRS (Connors Parent Rating Scale-Revised); SES (Socioeconomic status); BRIEF (Behavioral Rating Inventory of Executive Functioning); BRI (behavioral regulation index); MI (metacognition index);

* < .10;

* < .05;

** < .01