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Sleep Problems in Children With Autism Spectrum Disorder

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ABSTRACT OF THESIS

Sleep Problems in Children With Autism Spectrum Disorder

by

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The purpose of the current study was to examine the effect of restricted and repetitive behavior (RRB) on the relationship between sleep problems and externalizing behaviors in young children with autism spectrum disorder (ASD). Sixty-six participants with ASD and a mean age of 4 years and their parents participated in the study. Parents reported on their child's sleep, ASD symptoms, and externalizing behavior during a one-time lab visit. Bivariate correlations and a mediation analysis were conducted to assess the associations between sleep problems, externalizing behavior, and RRB. Results suggested that children with higher scores in measures of RRB had higher scores in sleep problems and externalizing behavior. Results also suggested that RRB partially mediated the relationship between sleep problems and externalizing behavior. Implications for the early identification of sleep and behavior problems, as well as ASD symptoms, are discussed. *Keywords:* restricted and repetitive behavior, sleep problems, externalizing behavior, autism spectrum disorder

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Introduction

Restricted and repetitive behavior (RRB) is a core characteristic of autism spectrum disorder (ASD), along with difficulties in social communication and interaction (American Psychiatric Association [APA], 2013). Though RRB is a defining feature of the ASD phenotype, the levels of severity and clinical presentation of specific RRBs are heterogeneous in nature (Georgiades, Szatmari, & Boyle, 2013; Hus, Pickles, Cook, Risi, & Lord, 2007; Kanner, 1943). As described in the Diagnostic Statistical Manual, fifth edition (DSM-V; APA, 2013), the criteria listed for RRBs entail a range of symptoms which include repetitive movements, repetitive use of objects and phrases, restricted interests, inflexibility to change in routines, ritualized patterns (e.g., insisting on listing items), and sensory seeking behaviors or adversity to sensory information in the environment (e.g., sensitivity to specific sounds).

Due to genetic and behavioral vulnerabilities, children with ASD often present with comorbid disorders and behavioral problems (Hollway et al., 2013; Mannion & Leader, 2013). Parents report that approximately 50-80% of children with ASD have sleep problems (Cortesi, Giannotti, Ivanenko, & Johnson, 2010; Couturier et al., 2005; Hundley, Shui, & Malow, 2016; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Mayes & Calhoun, 2009) including delayed sleep onset, shorter sleep duration, insomnia, and increased night-waking (Allik, Larsson, & Smedje, 2006; Couturier et al., 2005; Krakowiak et al., 2008; Mayes, Calhoun, Bixler, & Vgontzas, 2009; Souders et al., 2009). In contrast, prevalence rates of sleep problems among

neurotypical (TD) children are estimated to be between 20-30% (Sadeh, Mindell, Luedtke, & Wiegand, 2009).

In children with ASD, sleep problems may be related to increased risk for behavioral difficulties, which have an incidence rate ranging between 25-70% (Gregory & Sadeh, 2012; Hollway, Aman, & Butter, 2013; Sikora, Johnson, Clemons, & Katz, 2012). For example, externalizing behaviors (e.g., aggression, destruction of property, inattention) may be exacerbated by sleep problems (Gregory & Sadeh, 2012; Sikora et al., 2012). The converse direction of effect has also been suggested, such that more externalizing behaviors may worsen sleep problems in children with ASD (Abel et al. 2018; Park et al., 2015), although the direction of effects has yet to be conclusively determined. Though previous findings show a relationship between sleep problems and externalizing behaviors in children with ASD, there is little to no research examining how core ASD symptoms, particularly RRBs, influence that relationship, particularly in preschool children. Thus, the current study aimed to examine the effect of RRBs on the relationship between sleep problems and externalizing behaviors in young children with ASD.

Restricted and Repetitive Behavior

RRB can be separated into either low-level or high-level RRBs (Carcani-Rathwell, Rabe-Hasketh, & Santosh, 2006; Moore & Goodson, 2003; Prior & Macmillan, 1973; Turner, 1999). Low-level RRBs include motor behaviors such as hand flapping, body rocking, repetitive actions with objects (e.g., repeatedly flicking the eyes of a toy doll beyond initial object exploration) and stereotyped phrases or repeating vocalizations

(Boyd, McBee, Holtzclaw, Baranek, & Bodfish, 2009). On the other hand, high-level RRBs are behaviors such as resistance to change in routines, ritualized patterns of play, or intense preoccupations with a specific interest (Turner, 1999). Studies indicate that high-level RRBs are more common in older children with average to above average language and cognitive ability, while low-level RRBs are more common in children with below average cognitive ability and those who are preschool-aged (Militerni, Bravaccio, Falco, Fico, & Palermo, 2002.; Turner, 1999). Thus, these findings suggest that RRB may be related to developmental level. In addition, the presentation of RRBs can be a combination of low- and high- level behaviors which adds to the complexity of assessment and recognition (Leekam, Prior, & Uljarevic, 2011).

While RRBs are a core feature of ASD, they are not restricted to the disorder and often occur in TD children and in children with other types of neurodevelopmental disabilities (e.g., intellectual disability, Down syndrome; Evans et al., 1997; Evans & Gray, 2000). Restriction and repetition in behavior in early childhood can be typical of early development (Evans et al., 1997; Thelen, 1981). For example, it is not uncommon for a young TD child to insist on the same bedtime routine or to request to watch the same movie multiple times (Evans et al., 1997). Indeed, repetition of routines or behaviors have been posited as important tools for navigating and learning about the environment early in life (Evans et al., 1997; Thelen, 1981). Such routines often begin to decline after the age of three in TD children (McDonald et al., 2007); however, the same has not been shown for children on the autism spectrum (Kim & Lord, 2013).

To date, there is no clear consensus about whether or not RRBs decrease or increase with age among children with ASD. For example, some studies find that some RRBs, such as repetitive motor movements in preschool-aged children, become less prominent in middle childhood (South, Ozonoff, & McMahon, 2005). However, other studies find that RRBs, such as circumscribed interests, may increase in severity and frequency as children with ASD get older (Moore & Goodson, 2003; Bishop, Richler, & Lord, 2006; Richler, Bishop, Kleinke, & Lord, 2007; South, Ozonoff, & McMahon, 2005). More frequent and severe engagement of RRB in preschool-aged children may be linked to an ASD diagnosis later in development (Lord et al., 2006).

Sleep Problems and Restricted and Repetitive Behavior

There is evidence for a relationship between sleep problems and RRB in children with ASD (Hundley, Shui, & Malow, 2016; Tudor, Hoffman, & Sweeney, 2012; Verhoeff et al., 2018). According to the American Academy of Sleep Medicine (2014), sleep problems can be described as bedtime resistance, night awakening, insomnia, parasomnias, night terrors, daytime sleepiness, short sleep duration, and inconsistent sleep onset time. When compared to TD populations, children with ASD have significantly more sleep problems (Hirata et al., 2016; Matson, Ancona, & Wilkins, 2008). Additionally, children with ASD also experience more sleep problems relative to children with other types of disabilities like attention deficit/hyperactivity disorder (ADHD) or traumatic brain injury (Mayes et al., 2009). In terms of improvements in sleep problems for children with ASD, findings from a 4-year longitudinal study indicated that 31.5% of children with ASD experienced improvements in sleep problems

over time, whereas 21% experience worsening of sleep problems and 47.5% remain the same (Mazurek Dovgan, Neumeyer, & Malow, 2019). In contrast, the presence of sleep problems often decreases with age in TD children (Gregory & O'Connor, 2002; Sivertsen, Posserud, Gillberg, Lundevold, & Hysing, 2012).

With respect to the relationship between ASD symptoms and sleep problems, systematic literature reviews and cross-sectional studies suggest that higher scores on measures of RRB are significantly linked to more sleep problems (Adams, Matson, Cervantes, & Goldin, 2014; Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Hollway & Aman, 2011; Hundley, Shui, & Malow, 2016; Schreck, Mulick, & Smith, 2004). For instance, Gabriels et al. (2005) indicated that children with ASD and the presence of RRB on the Repetitive Behavior Scale (RBS; Lam & Aman, 2007) had higher scores for sleep problems on the Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirto, & McGuinn, 2000) relative to children with ASD without RRB. Similarly, parents reported that RRB, and particularly stereotypic behavior, was associated with fewer hours of sleep per night and increased night-wake screaming (Schreck, Mulick, & Smith, 2004). When children with ASD are placed into categories of "poor sleepers" and "good sleepers," findings suggested that poor sleepers had more ritualistic behavior when compared to good sleepers (Goldman et al., 2009). To date, it remains unclear how RRB relate to sleep problems in children with ASD, which suggests that further research is needed to understand this relationship.

Sleep Problems and Externalizing Behavior

In addition to RRB, the extant literature suggests that sleep problems in children with ASD can contribute to daily behavioral difficulties above and beyond the impact of ASD symptoms (Bradley, Summers, Wood, & Bryson, 2004; DeVincent, Kenneth, Gadow, Delosh, & Geller, 2007; Goldman et al., 2011; Gregory & O'Connor, 2002; Malow et al., 2006; Mayes & Calhoun, 2009; Sikora, Johnson, Clemons, & Katz, 2012). Children with ASD and sleep problems often exhibit more externalizing behaviors than children with ASD who do not experience sleep problems (DeVincent et al., 2007; Goldman et al., 2011; Gregory & Sadeh, 2012; Mayes & Calhoun, 2009; Sikora et al., 2012; Tureck, Matson, May, & Turygin, 2013).

For instance, children with ASD who slept fewer hours and had a delayed bedtime (i.e., stayed up later) had higher scores on measures of inattention, negative affect, aggressive behavior, and somatic complaints relative to good sleepers (Malow et al., 2006). In another study by Sikora et al. (2012), children with ASD were separated into groups consisting of children with moderate to severe sleep problems, children with mild sleep problems, and good sleepers. Results indicated that children in the moderate to severe sleep problems group had higher scores for externalizing behaviors (e.g., ADHD symptoms, aggression, oppositional defiant disorder) on the Child Behavior Checklist (CBCL; Achenbach, 2001), as well as lower scores on measures of adaptive functioning relative to children in the mild sleep problem and good sleepers groups (Sikora et al., 2012). Taken together, previous literature suggests that having severe sleep problems may increase the likelihood of having behavioral difficulties.

Among community samples of both children with and without ASD, having higher parent-reported scores on measures of ADHD symptoms and oppositional defiant behavior was linked to more sleep problems, such as trouble falling asleep, frequent night-waking, and crying at night (DeVincent et al., 2007). However, the authors found a greater percentage of sleep problems in the ASD group relative to the TD group (DeVincent et al., 2007). Though there are significant associations between sleep problems and externalizing behavior, less is known about the role of ASD symptoms, here RRB, in that relationship.

Restricted and Repetitive Behavior, Sleep Problems, and Externalizing Behavior

Links between RRB, sleep problems, and externalizing behavior are not well understood. In recent reviews by Cohen et al. (2014) and Hermann (2016), findings indicated that sleep problems were related to externalizing behaviors (e.g. aggression, attention) and ASD symptoms (RRBs). Goldman and colleagues (2011) examined sleep in the home setting using wrist actigraphy, which measures sleep duration, and found that children with ASD who were categorized as poor sleepers had higher scores on the CBCL for inattention and hyperactivity, as well as more RRBs than good sleepers. Likewise, children with ASD and sleep problems had elevated scores on the CBCL aggression and externalizing behaviors subscales when compared to children with ASD and no sleep problems (Park et al., 2012). Furthermore, Park et al. (2012) found that higher scores in RRB on the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & Le Couteur, 1994) were linked to insomnia in children with ASD. When examining the longitudinal relationship, sleep problems predicted subsequent frequency and severity of

RRBs, as well as negative affect and behavioral difficulties in 39 children with ASD between the ages of 2 to 10 years with ASD (Abel et al., 2018).

Furthermore, several studies suggest that sleep problems may exacerbate externalizing behavior, and externalizing behavior may worsen sleep problems over time (Abel et al., 2018; DeVincent et al., 2007; Gregory & O'Connor, 2002; Gregory & Sadeh, 2012; Hollway, Aman, & Butter, 2013; Maski & Kothare, 2013; Mayes & Calhoun, 2009). Findings from a longitudinal study with 490 TD children indicated that early behavioral problems, such as aggression and attention problems, predicted sleep problems in mid-adolescence (Gregory & O'Connor, 2002). Other studies have shown that sleep problems significantly predicted subsequent externalizing behavior in children with ASD (DeVincent et al., 2007; Hollway, Aman, & Butter, 2013; Mayes & Calhoun, 2009). Although RRB is a core symptom of ASD, to the investigator's knowledge, no published study has investigated how RRB influences the association between sleep problems and externalizing behavior.

Current Study

The current study examined the relationship between sleep problems, externalizing behaviors, and RRB in children with ASD and posed the following research questions: 1) What is the association between RRB, sleep problems, and externalizing behavior in children with ASD? 2) To what extent does RRB explain the relationship between sleep problems and externalizing behavior? It was hypothesized that there would be significant positive associations between RRB, sleep problems, and externalizing

behavior. It was also hypothesized that RRB would mediate the association between sleep problems and externalizing behavior.

Methods

Participants

A subset of 66 children with ASD (11F/59M; chronological age *M* = 4.05 years, *SD* = .98; range: 2.10-5-80) from a larger ASD screening clinic sample, participated in the study. Child race/ethnicity was based on an open-ended parent-report question and was then combined into the following classifications: Hispanic/Latino (45%), Other (26%), Caucasian (23%), Black (5%), and Asian (2%). Inclusion criteria for the selection of children for this study were a) had scores on measures of sleep, which was only collected for children between the ages of 1 to 6 years of age, b) confirmed ASD with the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000), c) meet for ASD given clinician judgement. Within the sample used for this study, 31% of participants were given a cognitive assessment using the *Wechsler Preschool and Primary Scale of Intelligence Second Edition* (WPPSI; Wechsler, 2012) or the *Differential Ability Scales Second Edition* (DAS-II; Elliot, 2006) while 67% were not able to be tested because they were either too young (under 2.5 years of age) or did not comply. The mean IQ was 66 (*SD*= 12.05; *range*=49-81).

The participants' mothers provided demographic information about themselves and their children. Thirty-seven percent of mothers reported that their highest education level was completing some school or had a high school diploma/GED. Another 37% of mothers reported having some college or an associate's degree/vocational diploma, while

27% reported having a bachelors or master's degree (0% of mothers reported having a doctorate or MD). In terms of household family income, most parents were either unsure or refused to answer (31%). Among parents who did report family household income, 28% earned under $25,000 a year before taxes, followed by 23% earning under $50,000 and 16% earning over $50,001. Eighty-three percent of parents reported English as the primary language spoken in the home, (15% Spanish; 5% other).

Measures

Participants were screened for ASD using the *Autism Diagnostic Observation Schedule Second Edition* (ADOS-2; Lord et al., 2012). The ADOS-2 is a standardized, play-based semi-structured observation that allows the assessor to directly observe the child's social and communicative behaviors. Children receive one of four modules based on their language ability (Toddler Module for children under 30 months of age, Module 1 for single or no words, Module 2 for phrase speech, and Module 3 for fluent speech). Scoring of the ADOS-2 is based on algorithms which provide summary scores for Social Affect (SA) and RRBs (Lord et al., 2012). Based on these scores, a child is placed into one of three categories: autism, autism spectrum, or non-spectrum. Lord et al. (2000) demonstrated that the ADOS has adequate to excellent test-retest reliability for individual items (κ = .59-82), interrater reliability (κ = .82-93), and internal consistency ($\alpha = .63 - .91$) for composite scores. Findings also indicated that the ADOS-2 has high specificity (α =.93–1.0) and sensitivity (α =.93–1.0) in differentiation between children with and without ASD. The ADOS-2 composite score derives a calibrated severity score (CSS;

Gotham, Pickles, & Lord, 2009), which ranges from 1-10, with 1 indicating low severity and 10 indicating higher severity. The CSS was used in this study.

Sleep problems and externalizing behaviors. Sleep problems and externalizing behaviors were measured using the *Child Behavior Checklist* (CBCL; Achenbach & Rescolar, 2001). The CBCL is a parent-report 100-item questionnaire used to measure symptoms of psychopathology; in this study, only the version for children between the ages of 1.5 to 5 years was used because it contains the sleep problem subscale. Parents were asked to describe their child's behavior within the past 6 months using a 3-point scale (0= *not true*, 1= *somewhat or sometimes true*, 2= *very true or often true*).

Six items comprise the sleep problems subscale; "sleeps less than most kids," "overtired," "trouble sleeping," "nightmares," "sleeps more than most kids during day and/or night," and "talks or walks in his/her sleep." The sleep subscale has been shown to have low to moderate internal consistency for (α=.48-.59; Goodnight et al., 2007; Gregory & O'Connor, 2002) which may be due to the fact that the scale is comprised of different kinds of sleep problems that are not particularly co-occur. The Externalizing Scale includes items assessing inattentiveness, hyperactivity, aggression, defiance, and noncompliance, and excludes the sleep scale items. Achenbach and Rescolar (2001) showed a high stability estimate of .87 for over an 8-day period, as well as reliability estimates of α=.89 for the Externalizing Scale. Total raw scores for both subscales are transformed into *t*-scores. *T*-scores at 69 or above indicate clinically significant symptoms, while *t*-scores between 65-68 indicate moderately clinical symptoms, and *t*-scores at or below 65 indicate behaviors in the average range.

Restricted and repetitive behavior. The *Social Responsiveness Scale Second Edition* (SRS-2; Constantino & Gruber, 2005) was used to measure RRB. The SRS is a parent-report 65-item questionnaire that assesses ASD symptoms. The SRS-2 provides scores for social communication and RRB (Constantino & Gruber, 2005). Parents rate ASD symptoms on a 4-point Likert scale from "1" (never true) to "4" (almost always true).

This study used the Restricted Interests and Repetitive Behavior subscale to capture RRB, which includes 12 items. The RRB subscale includes questions such as, "Has repetitive, odd behaviors, such as hand flapping or rocking." Summed scores are transformed to *t*-scores in which higher scores indicate greater symptom severity. *T*-scores are scaled to achieve equivalency for both female and male children (Constantino & Gruber, 2005). *T*-scores above 76 are within the severe range, scores between 66 to 75 are in the moderate range, while scores 74 or below are within the average range. Test-retest reliability of .88 after a three-month period has been shown among a clinic sample (Constantino & Gruber, 2005). Studies on the psychometric properties of the SRS have shown excellent internal consistency (α = .94 - .96) and test-retest reliability, as well as adequate inter-rater reliability ($\boldsymbol{r}$ = .61 - .92; Constantino & Gruber, 2012). Furthermore, excellent validity was found in content, construct, concurrent, and predictive validity (Constantino & Gruber, 2012).

Procedure

The Institutional Review Board of the University of California Riverside approved all study procedures. Parents and their children attended a one-time two-hour

visit to a university ASD screening clinic. Participants learned about the clinic from supporting agencies, recruitment events, flyers around the community, and referrals from educational or medical professionals. During the visit, parents were provided with informed consent in either English or Spanish by trained graduate students. Parents completed a demographic intake form and behavioral questionnaires while their child was given an ASD screening assessment and a measure of his or her cognitive ability. A demographic intake form was used to collect descriptive information about the child and parent.

Proposed Mediation Model

Bivariate correlations were conducted to examine the relationships between sleep problems, externalizing behavior, RRB, and demographic variables of interest (age, sex, maternal education, and household income, CSS). In addition to exploring the bivariate, mediation analyses were conducted in order to examine how RRB may contribute to the relationship between sleep problems and externalizing behavior. The rationale for the proposed mediation model was based on previous research suggesting that higher sleep problems may influence externalizing behavior via RRB (Goldman et al., 2011; Park et al., 2012). Thus, the question was to examine whether more sleep problems lead to more RRB which in turn lead to more externalizing behavior in children with ASD.

A priori power analyses using G*Power 3.1 (Faul, Erdfelder, Buchner, & Lang, 2009) suggested that a sample of 33 participants or more is sufficiently large to detect a large effect (f^2=. 33) with at least 80% power. The purpose for conducting a power

analysis was to examine whether a mediation analysis can be produced with the available sample of participants.

Before conducting the mediation analyses, the data were evaluated for violation of statistical assumptions for linear regression, such as linearity, homoscedasticity, normality, independence of residuals, and multicollinearity. Linearity was checked by plotting the studentized residuals against the unstandardized predicted values. The plot visually showed linearity between the dependent variable (externalizing behavior) and independent variables. In order to establish if a linear relationship exists between the dependent variable and each of the independent variables, partial regression plots were inspected which showed linear relationships across all independent variables. There was homoscedasticity, as assessed by visual inspection of studentized residuals versus unstandardized predicted values. Normality was checked by evaluating the statistics of the Kolmogorov-Smirnov and Shapiro-Wilk tests, in which both of the p-values were less than .05 thus violating normality. However, nonparametric bootstrapping procedures used when conducting a mediation analysis does not require a normal distribution (Hayes, 2013). There was independence of residuals, as assessed by the Durbin-Watson statistic of 1.99.

Based on Baron and Kenny (1986) a mediation analyses were conducted using the Statistical Package for Social Sciences (version 24; SPSS Inc., USA) with the PROCESS macro (Hayes, 2018). Step one examines whether sleep problems predict externalizing behavior (path c). Step two examines whether sleep problems predict RRB which is also known as path a. The last two steps evaluate whether both sleep problems and RRB

together predict externalizing behavior. Specifically, step three examines whether RRB predict externalizing behavior (path b) while step 4 examines whether sleep problems no longer or lessens the prediction of externalizing behavior (path c'). In order to assess the indirect effects of the mediation analysis, the non-parametric bootstrapping procedure using 5,000 samples, 95% bias correction and confidence intervals (CI) was implemented. For the indirect effect to be statistically significant, the CI should not cross zero (Hayes & Scharkow, 2013).

Results

Preliminary Findings

Descriptive statistics for all measures are shown in Table 1. For sleep problems, 58% of participants had scores in the average range, 33% in the clinically significant range, and 9% in the moderate range. For externalizing behavior, 52% of participants had scores in the average range, 43% in the clinically significant range, and 6% in the moderate range. For RRB, 52% of participants had scores in the severe range, 30% in the moderate range, and 18% in the average range.

Sleep Problems, Externalizing Behavior, and RRB: Correlations

For the first research question on exploring the relationship between RRB, sleep problems, and externalizing behavior, preliminary bivariate Pearson correlations were conducted along with demographic variables (sex, age, maternal education, household income) and CSS (Table 2). Results revealed significant positive correlations between sleep problems and externalizing behavior (r= .60, p<.001) and sleep problems and RRB (r= .37, p= .001). Significant positive correlations were also evident between

externalizing behavior and RRB (r= .50, p< .001). Externalizing behavior was negatively correlated with maternal education (r= -.37, p=.003), such that higher scores in externalizing behavior were associated with less maternal education. Also, externalizing behavior was positively correlated with child age, such that older participants had higher scores for externalizing behavior. Positive correlations between RRB and household income (r= .24, p=.05) were also found. The ADOS calibrated severity score (CSS) was not associated with RRB, sleep problems, or externalizing behaviors.

Maternal education was used as a covariate in the mediation given the its significant correlation with externalizing behavior and previous research supporting its impact on externalizing behavior (e.g., Silver, Measelle, Armstrong, & Essex, 2010). In addition, controlling for variables that are highly correlated is important, as they may bias the estimates of the mediation results (Valeri & VanderWeele, 2013). No other covariates were used in the final analyses given the lack of empirical support.

Sleep Problems and Externalizing Behavior: Mediation by RRB

For the second research question of to what extend does RRB explain the relationship between sleep problems and externalizing behavior, a mediation analysis was conducted with RRB mediating the relationship between sleep problems and externalizing behavior (Baron & Kenny, 1986). The Hayes (2018) PROCESS macro provides a guide of where the predictor variable goes in the model such that the predictor is inputted as (X), the mediator as (M), and the dependent variable as (Y). Thus, for the current study, the predictor variable was sleep problems (X), RRB as the mediator (M)

and externalizing behavior as the dependent variable (Y), while controlling for maternal education.

Results supported the suggested mediation pathway, see Figure 1. First, the regression of sleep problems on externalizing behavior, ignoring the mediator, was significant, (b=.58, SE=.09, 95% CI = [.00, .39], $p < .001$). Second, the regression of sleep problems on the mediator, RRB, was also significant (b=.45, SE=.14, 95% CI = [.17, .72], p=.002). Third, the mediation process indicated that RRB, while controlling for sleep problems, was significantly regressed on externalizing behavior, (b=.25, SE=.08., 95% CI = [.00, .09], $p < .001$). The completely standardized indirect effect was computed using 5,000 bootstrapped samples, with a 95% CI ranging from .02 to .23. The results also revealed a significant direct effect of sleep problems on externalizing behavior (b=.47, SE=.09, 95% CI = [.28, .66]). However, a partially standardized indirect effect was found (b=.01, SE=.00, 95% CI = [.00, .02]) suggesting that the relationship between sleep problems and externalizing behavior was lessened by RRB.

Discussion

The purpose of the study was to explore the relationship between RRB, sleep problems, and externalizing behaviors in children with ASD. In children with ASD, there are several studies suggesting that elevated scores on sleep problems measures are associated with elevated scores on measures of externalizing behavior (e.g., Gregory & Sadeh, 2012; Sikora et al., 2012). Given the complexity and range of ASD symptoms, there is limited empirical support on how ASD symptoms influence the relationship between sleep problems and externalizing behavior. Of those few studies, results show

that children with ASD experiencing more sleep problems have higher scores on measures of externalizing behavior and exhibit more RRB (Goldman et al., 2011; Park et al., 2012).

Thus, the first question aimed to examine the relationships between RRB, sleep problems, and externalizing behavior by conducting bivariate correlations. In accordance with previous studies, the results of the current study indicated that parents reporting higher levels of sleep problems also reported higher levels of RRB and externalizing behavior (Abel et al., 2018; Goldman et al., 2011; Park et al., 2012). Along with the significant positive correlations, demographic information was also included in the correlation matrix. Negative correlations were found between externalizing behavior and maternal education which suggests that more reported externalizing behavior was associated with lower maternal education. This finding is also consistent with previous literature showing that there is a link between socioeconomic status and behavioral difficulties among children (Silver, Measelle, Armstrong, & Essex, 2010).

Positive correlations were found between RRB and household income which suggested that higher household income was associate with more reported RRB. This finding has not empirical evidence, however further research should examine the ASD symptom reporting differences among parents from high and low socioeconomic backgrounds. Moreover, there was a positive association between child age and externalizing behavior which suggested that older children were reported to have more externalizing behavior. Mixed results have been found in regard to the decline or increase in externalizing behavior across age with some studies showing a decrease in behaviors

(e.g., Miner & Clarke-Steward, 2008), while other showed an increased (e.g., Goodnight et al., 2007).

The second question examined to what extent does RRB influence the relationship between sleep problems and externalizing behavior. Results indicated that RRB partially mediated the relationship between sleep problems and externalizing behavior, which suggests that RRB lessens the relationship between sleep problems and externalizing behavior but does not fully explain it. These findings are consistent with previous findings showing that there is a strong link between sleep problems and externalizing behavior (DeVincent et al., 2007; Gregory & O'Connor, 2002; Gregory & Sadeh, 2012; Hollway, Aman, & Butter, 2013; Maski & Kothare, 2013; Mayes & Calhoun, 2009); yet, what impact ASD symptoms have on this relationship has little empirical support (Goldman et al., 2011; Park et al., 2012). Although a full mediation was not found, this finding adds to the existing literature suggesting that RRB may exacerbate sleep problems and externalizing behavior in children with ASD (Goldman et al., 2011; Park et al., 2012).

Implications for Practice

Due to the increased prevalence of sleep problems and externalizing behavior in ASD, studies suggest that targeted interventions for sleep problems may improve daytime behavior. Potential strategies used to ameliorate sleep problems focus on principles of applied behavior analysis (ABA; Cooper, Heron, & Heward, 2007; Malow et al., 2012; Moon, Corkum, & Smith, 2011) and the use of medication (e.g., melatonin; Guénolé et al., 2011). Applied Behavior Analysis (ABA) strategies targeting sleep problems may

have the most empirical support and include: scheduled awakenings (Durand, Gernert-Dott, P., & Mapstone, 1996), bedtime fading with and without response cost (DeLeon, Fisher, & Marhefka, 2004; Sanberg, Kuhn, & Kennedy, 2018), consistent bedtime routine (Weiskop, Richdale, & Matthews, 2005), and parent education (Malow et al., 2014; Moon et al., 2011). Sanberg, Kuhn, and Kennedy (2018) used a bedtime fading strategy with response cost (BFRC; Piazza & Fisher, 1991) for reducing sleep problems among three children with ASD using a multiple baseline design. The bedtime fading strategies were comprised of adjusting the child's bedtime by 30 minutes each night depending on the sleep onset time of the previous night. For example, if a child went to bed within 15 minutes of bedtime, the bedtime for the following night was 30 minutes earlier. The response cost method consists of keeping the child awake by one hour if sleep was not initiated within the 15 minutes of bedtime onset time. Results showed a reduction in undesired co-sleeping a month after the bedtime fading strategy was implemented.

Another study used a parent education sleep curriculum to target sleep problems reported by parents on the Child Sleep Habits Questionnaire used lessons focused on a bedtime routine, strategies for bedtime resistance (e.g., gradually moving the chair closer to the door and out of the room, use of a bedtime social story), and homework in which parents kept track of strategies they used (Malow et al., 2014). The authors measured wrist actigraphy, a method used to measure for rest activity during sleep, and sleep diaries before and after implementing the sleep curriculum. Findings demonstrated positive gains in sleep habits using the actigraphy and a reduction of externalizing behavior based on parent-report measure. Similarly, Moon et al. (2011) used a parent

handbook called Better Nights, Better Days (Corkum et al., 2006) to reduce sleep problems and daytime behavior issues in three children with ASD. All three children had a reduction in delayed sleep onset difficulties and externalizing behavior. However, no changes in sleep duration were observed in the actigraphy data.

Lastly, other strategies include giving melatonin to children with ASD before bedtime. Melatonin is a hormone that is secreted in the pineal gland of the brain which aids the body's natural sleep cycle (Garstang & Wallis, 2006). A systematic review by Guénolé et al. (2011) suggests that melatonin improves sleep in children and adults with ASD. Although the findings above are promising for both behavioral strategies and medication, further research should examine the use of behavioral interventions based on ABA principles to target both sleep problems and externalizing behavior.

In addition, future research should examine best practices for targeting sleep problems and externalizing behavior early in development before the child starts school. The extant literature has shown that quality of sleep is significantly associated with academic performance and behavioral difficulties across development (Dewald, Meijer, Oort, Kerkhof, & Bogels, 2010). Particularly as young children adjust to school schedules, better sleep habits (e.g., more hours of sleep, early sleep onset) are associated with better test scores on reading and math (Bruni et al., 2006; Dewald et al., 2010; Perkinson-Gloor, Lemola, & Grob, 2013). Moreover, better sleep quality in later school grades (e.g., middle and high school) is associated with higher grade point average and better scores in math and reading (Wheaton, Chapman, & Croft, 2016).

Limitations and Future Directions

In light of present findings, several limitations should be acknowledged. First, the CBCL sleep subscale does not adequately capture all aspects of sleep problems in children. Several other measures, such as the Child Sleep Habits Questionnaire (e.g., Sikora et al., 2016) have been regularly used to examine sleep problems, whereas fewer studies have used the CBCL (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005). In the current study, sleep problems were based on parent report, which is likely not as accurate as objective measures (e.g., wrist actigraphy). Future research should include alternative or additional measures such as a sleep log or wrist actigraphy along with parent-report questionnaires.

Second, the RRB subscale of the SRS may not accurately represent the heterogeneous nature of RRBs. Previous literature investigating RRB in youth with ASD used measures specifically targeting a large number of RRBs (e.g., RBS-R; Bodfish, Symons, & Lewis, 1999). Therefore, future research should investigate the mediating effect of RRBs using a measure that captures a wider variety of RRBs. In order to disaggregate the variable of RRB, future research should also investigate the differences between low-level and high-level RRBs among a diverse sample of children with ASD. Third, this study only looked at children with ASD and did not include a sample of neurotypically developing (TD) children.

Lastly, the data analysis method may not have captured the effect of RRB on the relationship between sleep problems and externalizing behavior. For instance, the normality assumption for regression was violated which may suggest that the data was

not normally distributed. However, as noted previously, bootstrapping procedures employed by the PROCESS macro on SPSS is a nonparametric procedure which creates a normal distribution based on the sample data (Hayes, 2013).

Conclusion

The results of this study contribute to the growing literature suggesting RRB as a potential mediator in the relationship between sleep problems and externalizing behavior for young children with ASD. Furthermore, these findings support evidence suggesting that sleep problems and externalizing behavior are interrelated. Young children with ASD are at heightened risk for comorbid symptoms of psychopathology, which often emerge in early childhood and persist over time (Burt & Roisman, 2010; Humphreys & Zeanah, 2015; Pihlakoski et al., 2006). Given the elevated prevalence rates of sleep problems and externalizing behavior among children with ASD, clinicians and parents may wish to target these difficulties in order to reduce the frequency of these behaviors, and their potential exacerbation, before the child starts school. For a child with ASD, increased sleep problems and externalizing behaviors may impede academic success (Sikora et al., 2012) and negatively influence peer relationships (Kasari, Locke, Gulsrud, & Rotheram-Fuller, 2011). It is paramount for professionals such as school psychologists to be aware of the relationship between sleep problems and externalizing behavior in children with ASD in order to develop effective treatment strategies.

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Table 1

Means, standard deviations, and ranges for sleep problems, externalizing behavior, RRB and demographic variables of interests.

Variable	Mean (SD)	Range
Sleep Problems	66.26 (13.98)	50-100
Externalizing Behaviors	66.65 (14.07)	39-97
RRB	78.08 (16.46)	42-103
CSS	6.13 (2.05)	1-10
Child Age in Years	4.09 (1.04)	2.10- 6.30

Note: RRB= Restricted and Repetitive Behavior. CSS= Calibrated Severity Score

Table 2

Correlation matrix with sleep problems, externalizing behavior, RRB and demographic variables.

Variable	1	2	3	4	5	6	7	8
Sleep Problems	--							
Externalizing Behavior	.60**	--						
RRB	.37**	.50**	--					
CSS	-.16	-.15	.12	--				
Child Age	.13	.24*	.02	-.16	--			
Child Sex	-.14	-.00	.03	.07	.06	--		
Maternal Education	-.09	-.37*	-.11	.12	-.05	.00	--	
Household Income	.02	-.04	.24*	.01	.11	.02	.20	--

Note: RRBs= Restricted and Repetitive Behaviors. CSS= Calibrated Severity Score. **$p < .001$, *$p < .05$

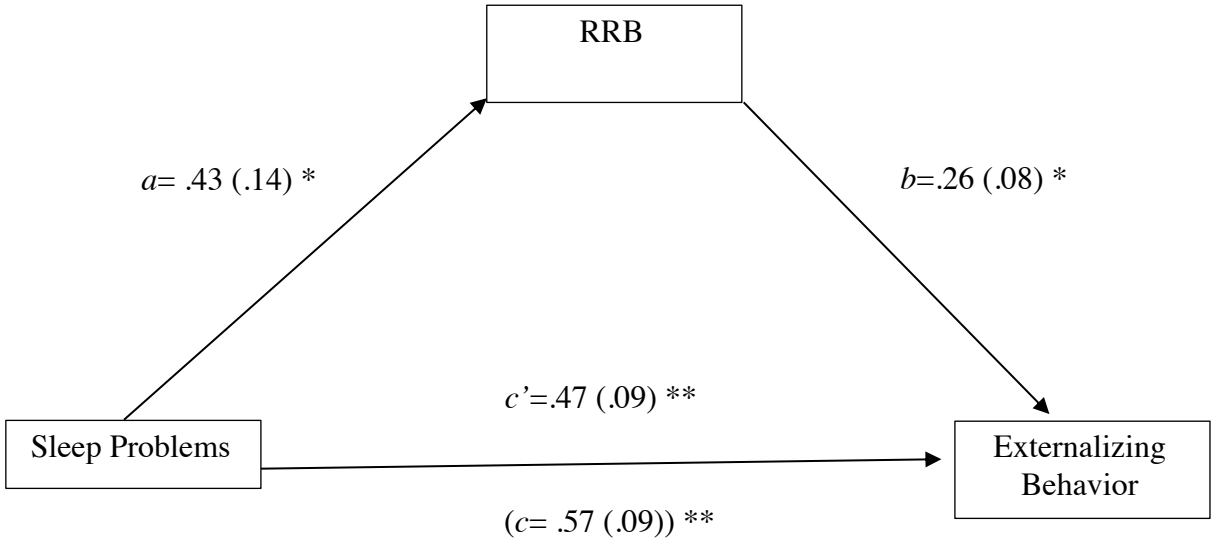

Figure 1. Standardized regression coefficients for the relationship between sleep problems (independent variable) and externalizing behavior (dependent variable) as mediated by RRB, while controlling for maternal education. Note. Coefficient (standard error). RRB= Restricted and Repetitive Behavior. * $p < .05$, ** $p < .001$