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Exploring Sleep Quality of Young Children with Autism Spectrum Disorder and Disruptive Behaviors

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Abstract

Background and purpose—Sleep disturbances in autism spectrum disorder (ASD) are common and may impair daytime functioning and add to parental burden. In this well characterized sample of young children with ASD and disruptive behaviors, we examine the association of age and IQ in sleep disturbances using the Child Sleep Habits Questionnaire modified for ASD (CSHQ-ASD). We also test whether children with poor sleep have greater daytime behavioral problems than those with better sleep. Finally, we examine whether parental stress is higher in children with greater disruptive behaviors and sleep disturbances.

Participants and methods—Participants were 177 with complete data of 180 children (mean age 4.7) with ASD who had participated in a randomized clinical trial. Parents completed the CSHQ-ASD and several other measures at study enrollment. The sample was divided into “poor sleepers” (upper quartile on the total score of the CSHQ-ASD) and “good sleepers” (lower quartile) for comparisons. Analyses were conducted to evaluate group differences on age, IQ, daytime disruptive behavior, social disability and parental stress.

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Results—The two groups of young children with ASD, good sleepers versus poor sleepers, were not different on age or cognitive level. Children in the poor sleeping group had significantly higher daytime behavioral problems including irritability, hyperactivity, social withdrawal and stereotypical behaviors. Parents in this group reported significantly higher levels of stress.

Conclusions—The finding of no age difference between good and poor sleepers in young children with ASD and disruptive behaviors suggests that sleep problems are unlikely to resolve as might be expected in typically developing children. Likewise, the good and poor sleepers did not significantly differ in IQ. These findings add strong support for the need to screen for sleep disturbances in all children with ASD, regardless of age and cognitive level. Poor sleepers exhibited significantly greater daytime behavioral problems and parents of children in this group reported significantly higher levels of stress. Above and beyond the co-occurring disruptive behavior, poor sleep quality appears to pose substantial additive burden on child and parents..

Introduction

Autism Spectrum Disorder (ASD) is characterized by social communication deficits, restrictive interests and repetitive behavior (1) affecting 6.2 – 14.7 per 1000 children depending on study design and sample ascertainment (2, 3). In addition to these core diagnostic features, many children with ASD have co-occurring problems such as tantrums, aggression, self-injury, hyperactivity, anxiety, depression, and sleep disturbance (4–9). Although the etiological connections are not clear, children with ASD may have one or more of these concomitant conditions. Sleep disturbances such as bedtime resistance, sleep-onset association problems, delayed sleep onset, interrupted sleep and decreased total sleep time have been reported in up to 80% of children with ASD (8–16).

The trajectory of sleep problems in typically developing (TD) children tends to be worst in the early preschool years and diminish over time(17). Although findings are not completely uniform, age is not strongly related to sleep disturbances in children with ASD (18–21). More recently, in a sample of 3–17 year olds, sleep difficulties were highest at ages 6–9 for children with ASD during which time sleep disturbances had greatly decreased in TD children (22). In one of the first studies to examine sleep longitudinally, sleep disturbances emerged after 18 months and persisted through 11 years of age in children diagnostic with ASD (23). This was in sharp contrast to the TD children in the study sample whose sleep problems resolved in the preschool years.

In children with ASD, cognitive functioning has been proposed to play a role in sleep problems, but the relationship is unresolved with studies offering mixed results. Although some studies report that sleep problems are more common in children with ASD and lower cognitive abilities (20, 24–28) other studies have not confirmed this association (8, 14, 18, 19). Despite the inconsistencies, it is clear that sleep disturbances in children with ASD occur across cognitive levels.

A growing literature supports links between poor sleep and behavioral and emotional problems in children with ASD. Inattention, hyperactivity, aggression, self-injury and anxiety are consistently more common in children with ASD and diminished sleep quality (16, 29–32).

Finally, sleep difficulties in children with ASD may impose additional stress on caregivers and foster negative attitudes toward the child (28, 33–35). Parents of children with ASD have lower sleep quality than parents of typically developing children (36, 37), especially when the child with ASD has sleep problems (38). Sleep problems are also associated with maternal depressive symptoms (39).

The current study aimed to extend previous work in the area where findings thus far have been equivocal, in a well characterized sample of young children with ASD and moderate to severe disruptive behaviors. In this sample, we examine the role of age and IQ in sleep disturbances. We also test whether children with poor sleep have greater daytime behavioral problems and whether parental stress is greater in children with sleep disturbances compared to those with ASD and adequate sleep.

Methods

Sample

The sample consisted of 177 of the 180 children who participated in the Research Units on Behavioral Intervention (RUBI) study (three participants had incomplete sleep data). RUBI was a 24-week randomized controlled trial (RCT) of children ages 3–7 years of age who were randomized to Parent Training (PT) or a Parent Education Program (PEP) (40, 41). Eligible children were healthy, had a diagnosis of ASD, and moderate or greater severity of disruptive as evidenced by a score of ≥ 15 on the Aberrant Behavior Checklist (ABC) and a Clinical Global Impressions Severity (CGI-S) score of ≥ 4 . Participants were on no medication or on stable medication for at least 6 weeks and no planned changes for six months were included. Diagnoses of Rett Disorder or Childhood Disintegrative Disorder, any serious medical conditions or psychiatric disorder, developmental level of less than 18 months and current or past enrollment in structured PT program were study exclusions. The study was initiated prior to DSM-V (1). Thus, the ASD diagnoses were based on DSM-IV criteria (42), corroborated by the Autism Diagnostic Observation Schedule (ADOS) (43, 44) and Autism Diagnostic Interview – Revised (45). This six study sites included the Emory University, Indiana University, Ohio State University, University of Pittsburgh, University of Rochester and Yale University.

Study Measures

Demographics—Parents completed a demographic form at study entry.

Developmental/Cognitive functioning—The Abbreviated Battery of the Stanford-Binet Intelligence Scales: 5th Edition(46) or the Mullen Scales of Early Learning(47) were used to assess each participant's level of intellectual functioning. Most children were assessed with the Stanford-Binet. The Mullen was used for children with limited language skills, mental age below 3 years or children who were unable to complete the Stanford-Binet.

Children's Sleep Habits Questionnaire (CSHQ)(48) is a 33-item, parent-report measure designed to screen for sleep problems in children aged 4 to 10. A subsequent study supported the validity of the CSHQ in children as young as two years of age (49). The

CSHQ items are rated on a 3-point scale: 3 = usually (5–7 nights per week); 2 = sometimes (2–4 nights per week); and 1 = never/rarely (0–1 nights per week). In addition to the frequency ratings, a ‘yes’ or ‘no’ response to the question “is this a problem?” is included. The summation of the frequency ratings creates the total score, with higher scores reflecting greater sleep disturbances. The original CSHQ has 8 subscales: bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing, and daytime sleepiness. These subscales were based on the pediatric International Classification of Sleep Disorder (48) and subsequent factor analyses have not confirmed these subscales psychometrically (50–52). Although the CSHQ was not developed for children with ASD, it has been widely used to characterize sleep problems in this population. Recently, we explored the psychometric properties of the CSHQ in a relatively large sample of 310 children with ASD (53). The revised measure includes 27-items (six items did not load on any factor) with five components. The subscales were labeled: 1) Sleep Routine Problems (9 items); 2) Insufficient Sleep (5 items); 3) Sleep-Onset Association Problems (4 items); 4) Parasomnia/Sleep Disordered Breathing (6 items); and 5) Sleep Anxiety (3 items). This version of the CSHQ, here after called the CSHQ-ASD, was used in this study. For purposes of this study, we used the total sleep score of the 27-item revised measure.

Aberrant Behavior Checklist (ABC)(54) is a 58-item parent rating scale rated on a four-point scale from 0 (not a problem) to 3 (severe in degree). The ABC contains five subscales: Irritability (15 items), Social Withdrawal (16 items), Stereotypic Behavior (7 items), Hyperactivity/Noncompliance (16 items), and Inappropriate Speech (4 items). The ABC has been validated in children with ASD(55). The Irritability, Hyperactivity, and Social Withdrawal subscales have been used as outcomes for several RCTs in ASD(40, 56–58). Here we were interested in the Irritability and Hyperactivity/noncompliance subscales as indices of disruptive behavior, Social Withdrawal subscale as a measure of social disability, and Stereotypy as an index of repetitive movements.

Parenting Stress Index- Short Form (PSI)(59) is a 36-item measure completed by parents of children 3 months to 10 years of age designed to assess parental stress. Each item is rated on a 5-point scale. The PSI includes three factors: Parental Distress, Parent-Child Dysfunctional Interaction, and Difficult Child Characteristics. Example statements include, “I feel trapped by my responsibilities as a parent,” “Sometimes I feel my child doesn’t like me and doesn’t want to be close to me,” and “I feel that my child is very moody and easily upset.” The PSI has good test-retest reliability (.96) and internal consistency (.90). A total score of 88 (85th percentile) and above is considered in the clinically significant range for parental stress. This measure has been shown to capture parental stress associated with raising a child with ASD and disruptive behavior. The PSI provides insight into parents’ internal emotional state, as well as the stress related to child behavior and parent-child interactions(60).

Data Analyses

Data analyses were conducted using PASW statistics (Version 22)(61). We divided the sample into “poor sleepers” (upper quartile on the total score of the CSHQ-ASD) and “good sleepers” (lower quartile) for comparisons. Descriptive statistics were calculated for the

entire sample as well as the two groups (poor sleepers; good sleepers). Chi Square test was performed to determine group differences in IQ (dichotomous variable of 70 and above, lower than 70).. T-tests were conducted to evaluate group differences on age, daytime disruptive behavior and parental stress.

Results

Demographic information is provided in Table 1. This sample was mostly Caucasian, non-Hispanic and parents were relatively well educated. The mean age of the study sample (N=177) was 4.7 ± 1.14 years. There were no age differences between the good sleepers (n= 52 with a CSHQ-ASD score in lower quartile) and poor sleepers (n= 46 with a CSHQ-ASD score in upper quartile) ($t=0.58$; $p = 0.57$). In both good and poor sleep groups, over 70% of participants had an IQ of 70 or above, and there was no significant difference in good sleepers compared to poor sleeper on this dichotomous IQ variable (>70 ; <70) ($\chi^2=0.03$, $p = 0.87$). In contrast, poor sleepers had significantly higher scores on the ABC Irritability, Hyperactivity, Stereotypy and Social Withdrawal subscales. Likewise, all the subscales of the PSI and the PSI total score were significantly higher in the poor sleeper group compared to children in the good sleeper group.

Discussion

This study examined sleep in a sample of young children (age 3 to 7 years) with ASD with moderate to severe disruptive behavior. The well characterized sample included participants from a randomized trial of parent training versus parent education (40, 41). Pre-treatment primary caregivers (mothers in most cases) completed numerous paper and pencil measures at study enrollment. For this study we stratified the sample in order to compare children who had sleep disturbances to children who did not. Sleep disturbance was measured on the 27-item CSHQ-ASD. This modified version has fewer items than the original 33-item CSHQ and has a different factor structure.(53).

We found no evidence of an association between age and sleep disturbance in our sample. This finding should be interpreted with caution in view of the restricted age range of 3–7 years. However, this range does include the period in which TD children show the sharpest decline in sleep problems(23). Because our finding suggests that sleep problems are unlikely to resolve as might be expected in similar ages of TD children, early treatment for sleep disturbances for young children with ASD may be warranted (9). Likewise, the good and poor sleepers did not significantly differ in IQ. Collectively, these findings add support the value of screening for sleep disturbances in all children with ASD, regardless of age and cognitive level.

Although significant differences were not observed for age or cognitive functioning, poor sleepers had significantly higher mean scores on four of five ABC subscales (Irritability, Hyperactivity, Social Withdrawal, and Stereotypy). The largest differences in children with poor sleep were in irritability and hyperactivity/noncompliance, suggesting a possible connection between poor sleep and degradation in externalizing, daytime behaviors. This relationship between sleep behaviors and similar externalizing behaviors has been reported

previously in primarily correlation studies (16, 30, 32, 33, 62). Our cross section design does not allow determination of the direction of this relationship, but either direction is plausible and has been suggested in the general pediatric literature for children without ASD (63, 64). A child who sleeps poorly is likely to be more irritable and hyperactive, but conversely children who are irritable and hyperactive may not settle for sleep. Further inquiry is called for with longitudinal study designs to disentangle the likely bidirectional associations and their magnitude (63, 65). Given our findings here and previous work, clinicians providing empirically supported parent training interventions for disruptive behavior in children with ASD should also consider integrating sleep interventions in the treatment plan(40).

In addition to scores on disruptive behavior measures, the Social Withdrawal and Stereotypy subscales on the ABC were significantly different for the two groups with higher scores for the poor sleepers. The Social Withdrawal subscale is a reliable indicator of social disability in children with ASD and is sensitive to change with treatment (57). This finding suggests those children with poorer sleep display more social disability, but again causality or direction of this interaction is not clear. Higher scores on the ABC Stereotypy subscale in the poor sleepers also suggests an interaction. Although measured on different instruments, the increase in stereotypical behaviors is similar to other reports of increased repetitive and ritualistic behaviors in children with ASD and sleep problems (25, 30). In a prior sleep treatment study, we speculated that greater stereotypy was related to poorer treatment response, but the sample size was insufficient to examine this possibility (9). Again, further studies with designs that will allow examination of the complex interactions between sleep, social disability and repetitive behaviors are needed.

Along with the significantly higher daytime behavior problems in the poor sleep group, parenting stress was pointedly higher as well. All subscales of the PSI were significantly greater in the poor sleeper group compared to the good sleeper group. This indicates that parents face greater burden when a child with ASD has the combined clinical picture of disruptive behavior and poor sleep patterns. As noted, the potentially bidirectional nature of these associations deserves more investigation in the field of ASD. It may be that poor child sleep increases parental stress; just as plausible is that parent stress and poor parent-child interactions unsettle the child's sleep. These complex, transactional relationships between parent-child interactions around bedtime and nighttime have been examined in young children with typical development (66–68), but not in children with ASD. These relationships should be studied within a biopsychosocial context as proposed by Sadeh and colleagues (69, 70). Understanding these relationships is particularly important for treatment development as it has been shown that parenting practices can be modified to benefit their children with ASD in other areas (40, 41, 71–73).

This study has several limitations that warrant consideration. First, our sample of children with ASD included a narrow age range from age 3 to 7 years. To be included in the study, all children had to have at least moderate levels of disruptive behaviors. This requirement limits the generalizability of the findings. Although IQ did not seem related to sleep disturbances in this sample, there is a chance that we underestimated IQ given the verbal requirements of the two measures used the IQ tests used (74). Moreover, 87% of our sample was Caucasian, which may not be representative of all young children with ASD and disruptive behaviors.

Nonetheless, our findings support an interaction between sleep disturbances, disruptive behavior, social disability, repetitive behaviors, and parental stress in young children with ASD. These documented associations indicate the need for further study on the underlying multifaceted mechanisms related to develop of sleep patterns in children with ASD.

Based on our findings here and the extant literature suggesting that poor sleep is linked to a variety of daytime behavior problems and parent stress but possibly not to age or cognitive level, further study of sleep within a comprehensive biopsychosocial model is needed to advance our understanding of why some young children with ASD have significantly disrupted sleep patterns and others do not is warranted. Such studies could examine the relative contributions of child biomarkers to include cortisol and serotonin given reported abnormalities in ASD which may affect circadian rhythms (75–78). Linkages between frequently co-occurring psychiatric conditions in ASD such as anxiety or attention problem and sleep patterns should be explored in longitudinal studies to move beyond correlation. Despite the demonstrated contributions of parent and family variables in disrupted in sleep patterns in typically developing children, parental behavior at bedtime and interrupted sleep patterns of young children with ASD are unexplored (66, 68, 79, 80). The role of other family characteristics in sleep disturbances in children such as socioeconomic status and family composition; which have studied in other pediatric populations have not been examined in ASD. Learning more about these complex interactions of biological and environmental factors will promote development of more tailored interventions to improve sleep quality in children with ASD. With improved sleep, better outcome for children with ASD could be expected.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Highlights

- No age difference between good and poor sleepers in young children with ASD and disruptive behaviors were seen. This suggests that sleep problems are unlikely to resolve as might be expected in similar ages of children with typical development.
- Likewise, the good and poor sleepers did not significantly differ in IQ. These findings add strong support for the need to screen for sleep disturbances in all children with ASD, regardless of age and cognitive level.
- In contrast, poor sleepers exhibited significant more daytime behaviors to include irritability hyperactivity, social withdrawal and stereotypical behaviors
- Parents of the poor sleep group experienced significantly more stress.

Table 1

Descriptive Characteristics of Study Sample

	Total Sample N=177	Good Sleepers N= 52	Poor Sleepers N =46	p value
Mean (SD) age (in years)	4.7 (1.14)	4.81 (1.07)	4.67 (1.23)	0.57
Intellectual Level^a	# (%)	# (%)	# (%)	
70	132 (74.6)	40 (76.9)	34 (73.9)	0.87
<70	43 (24.3)	12 (23.1)	11 (23.9)	
Gender				
Male	155 (87.6)	46 (88.5)	38 (82.6)	
Female	22 (12.4)	6 (11.5)	8 (17.4)	
Diagnosis				
Asperger	5 (2.8)	3 (5.7)	0 (0)	
Autism	116 (65.5)	36 (69.2)	26 (56.5)	
PDD	50 (28.2)	13 (25)	17 (37)	
Race/Ethnicity				
Caucasian/White	154 (87)	46 (88.5)	40 (86.9)	
African American	14 (7.9)	2 (3.8)	5 (10.9)	
Asian	6 (3.4)	3 (5.8)	3 (3.1)	
Pacific Islander	2 (1.1)	0 (0)	1 (2.2)	
Other/Mixed	1 (.6)	1 (1.9)	0 (0)	
Hispanic	26 (14.7)	5 (9.6)	11 (23.9)	
Non-Hispanic	151 (85.3)	47 (90.4)	35 (76.1)	
Maternal Education				
Advanced degree	51 (28.8)	17 (32.7)	12 (26.1)	
College degree	57 (32.2)	21 (40.4)	11 (23.9)	
Some College	54 (30.5)	12 (23.1)	17 (37.0)	
High school	7 (7.9)	2 (3.8)	5 (10.9)	
Some high school	1 (.6)	0 (0.0)	1 (2.2)	
ASD CSHQ	Mean (SD)	Mean (SD)	Mean (SD)	
Sleep Routine Problems	13.81 (3.60)	10.85 (1.79)	17.91 (2.76)	
Insufficient Sleep	7.58 (2.61)	5.92 (1.15)	9.13 (3.04)	
Sleep-onset Association Problems	6.4 (2.93)	4.63 (1.36)	9.46 (2.66)	
SDB/Parasomnias	9.02(2.37)	7.29 (1.53)	11.24 (2.29)	
Sleep Anxiety	4.12 (1.22)	3.42 (.67)	4.93 (1.34)	
Total	40.94 (8.62)	32.12 (2.49)	52.67 (5.08)	

^a 2 missing

ABC & PSI Results

Table 2

	Total Sample N=177		Good Sleepers N=52		Poor Sleepers N=46		t- statistic	p value
	Mean (SD)		Mean (SD) ^a		Mean (SD)			
ADOS Total	15.09 (4.57)		14.75 (4.46)		15.07 (4.33)		0.36	.72
ABC Subscales								
Irritability	23.80 (6.29)		22.12 (5.34)		25.83 (7.25)		2.85	.005 *
Social Withdrawal/Lethargy	12.87 (8.22)		11.81 (7.32)		15.59 (8.52)		2.34	.021 *
Stereotypic Behavior	6.39 (4.92)		6.08 (4.79)		8.11 (4.73)		2.11	.038 *
Hyperactivity/Noncompliance	30.34 (9.32)		28.13 (9.34)		33.96 (9.13)		3.11	.002 *
Inappropriate Speech	5.75 (3.15)		5.33 (3.22)		6.52 (3.13)		1.86	.066
PSI Subscales	Mean (SD)	%ile	Mean (SD)	%ile^a	Mean (SD)	%ile^b		
Parental Distress	32.95 (8.72)	74	30.22 (8.34)	65	35.69 (9.72)	79	2.91	.005 *
Parent-Child Dysfunctional Interaction	28.85 (7.49)	83	26.31 (5.77)	79	31.76 (7.39)	89	3.96	.000 *
Difficult Child	42.55 (7.54)	94	40.63 (7.25)	91	44.98 (9.13)	97	2.54	.013 *
Total Stress	104.13 (20.04)	91	97.16 (17.27)	87	111.53 (24.99)	95	3.22	.002 *

^a 3 PSI missing^b 1 PSI missing

* Significant