SLEEP AND COGNITION
IN INDIVIDUALS WITH AUTISM AND DOWN SYNDROME

BY

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Acknowledgments

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Abstract

Individuals with Autism (ASD) and Down syndrome (DS) have poor sleep (Churchill et al., 2013; Lambert, Tessier, Chevrier, Scherzer, Mottron, and Godbout, 2013; Wiggs and Stores, 2004). However, the type of poor sleep differs between groups. We wanted to explore the nature of parent reported sleep differences in autism and Down syndrome, and compare sleep severity to verbal and nonverbal IQ. We aimed to unveil the relation between sleep and cognition between both groups. Both populations (N=36) were given the Child Sleep Habits Questionnaire (CSHQ), a parent-reported questionnaire, and were tested on IQ by the Kaufman Brief Intelligence Test-II (KBIT-II). Our results yielded significantly higher scores for sleep onset delay and daytime sleepiness in ASD, and a marginally significant increase in bedtime resistance for DS. We compared the 9 subscales of the CSHQ to verbal and nonverbal IQ. Our results showed a significant correlation for ASD between the total sleep disturbance score (TSDS) and verbal IQ. As a more reliable measure of sleep, the CSHQ would be better utilized in conjunction with a more objective measure of sleep, such as polysomnography (PSG).

Keywords: Autism Spectrum Disorders, cognition, CSHQ, Down syndrome, sleep
In the literature on ASD, sleep is not fully understood in conjunction with memory, behavior, and cognition. Sleep problems are thought to exacerbate repetitive and stereotyped behavior. These individuals have reported high frequency night awakenings, parasomnias, and tiredness during the day compared to the control group (Miano et al., 2007). There are various factors that are attributed to sleep problems in ASD. These include: reported insomnia, poor sleep hygiene, repetitive behaviors, and brightly lit screens in bedroom, which is thought to disrupt melatonin production and was significantly related to less sleep (Adams, Matson, Cervantes, & Goldin, 2014; Engelhardt, Mazurek and Sohl, 2013; Malow et al., 2009).

ASD is characterized as a spectrum disorder because of the range of severity and mannerisms. Individuals with ASD have difficulty with social and communicative behaviors, as well as restricted interests and repetitive behaviors (DSM-5, 2013). Hollway, Aman and Butter (2013) researched other factors that could be considered “risk markers” resulting from disturbed sleep in individuals with autism. They explored factors that are possibly affected in ASD: cognitive, behavioral, emotional, and physiological. They created a theoretical framework to predict disturbed sleep. The bidirectional theoretical framework looks at intellectual functioning as a moderator for the severity of autism, linking this variable to the measure of disturbed sleep. People with ASD that have a pre-existing “vulnerability”, in conjunction with environmental stressors, could develop sleep disturbances. Their research has shown that comorbidity of other medical conditions, in part with lack of sleep, creates maladaptation social responses. They found a positive correlation between IQ and Sleep Anxiety Outcomes (e.g. sleep onset delay, bedtime resistance, sleep disordered breathing, parasomnias, and total sleep disturbance score) based on the Child Sleep Habits Questionnaire, CSHQ (Hollway et al., 2013). There was a positive correlation between cognition and sleep anxiety. This demonstrates that it might be
necessary, but not substantial, when considering “vulnerability factors”, to consider level of intellectual functioning alone (Hollway et al., 2013). They concluded that the strongest predictor of sleep disturbance that they found was anxiety.

The nature of sleep disruption in ASD has been researched and compared to typically developing individuals (TD), and individuals with DS. Aihara & Hoshimoto (1986) conducted a sleep study using PSG. Results showed abnormal REM activity (spindling-like), and during stage 1 and 2 of NREM sleep, they saw a presence of rapid eye movements. These findings suggest that there is maturational process defect in ASD. They did report that sleep abnormalities tend to normalize across development for people with ASD (as seen in Diomedi et al., 1999). The literature also shows that REM latency is reportedly shorter for participants with Autism. When comparing ASD to DS, ASD had a significantly higher amount of REM sleep and activity. However, compared to TD, ASD showed a significant reduction in the percentage amount of REM sleep. Malow et al. (2009), developed a measurement that assess’ sleep hygiene in children with ASD, the Family Inventory of Sleep Habits. Items of the inventory “were generated to ascertain daytime behaviors known to influence sleep that would also be amenable to change with treatment” (p.20). The parents are asked to indicate the frequency of sleep habits based on a Likert scale of 1-5 (1-never, and 5-always). Higher scores are indicative of more typical sleep habits. The findings of this study suggest that the instrument is positively correlated with the Peabody Picture Vocabulary Test-III, which supports their hypothesis that higher vocabulary knowledge is associated with better sleep hygiene. The results suggest knowledge (conceptual, verbal, nonverbal: based on vocabulary knowledge) may be associated with sleep. Therefore, it is imperative to test different facets of knowledge, as well as specific factors of sleep.
Individuals with ASD have significant sleep disturbances that are also reported in other neurodevelopmental disorders (NDD), such as Down syndrome. Down syndrome is used as a comparison group because it provides an alternative model of disrupted sleep. While both populations have low quality sleep, they differ in the context of poor sleep, as well as undergo different developmental trajectories. Sleep disturbances are as common in autism as they are in children with DS (Richdale, 2000). Though there was no exact sleep measure that correlated with disruptive behavior throughout the day, energetic daytime behaviors were related to sleep trouble. When compared, significant correlations were found between sleep problems and daytime behavior (Patzold et al., 1998; as seen in Richdale, 2000). These findings suggest that there is a correlation between behavior and sleep, not only in DS, but in autism as well. DS is largely affected by Obstructive Sleep Apnea Syndrome (OSAS) (Breslin et al., 2011), as determined by PSG testing. While ASD is primarily affected by sleep onset delay. When assessing different variables within DS and ASD, it is important to consider disrupted sleep as an inevitable factor of having either disorder. Sleep problems could be related to underlying problems that are exacerbating poor sleep, such as anxiety, social and communication skill deficits, and other delayed variables. However, this relationship should be further investigated (Richdale, 2000).

Sleep, behavior, and cognition vary in development among different NDDs. Though sleep problems are prevalent in ASD and DS, the exact nature of these problems differ between both groups. For instance, OSAS can be observed in individuals with DS as young as 2-4 years old. Apneas occur intermittently throughout the sleep period, and are a product of obstruction in the airway caused by relaxed throat muscles. When the airway is blocked, the individual cannot breathe and consequently awakens. Since these episodes happen frequently, individuals with
OSAS have poor quality sleep due to many night awakenings. This prevents the individual from consolidating new material from the day, as we consolidate memories and vocabulary during sleep (Walker & Stickgold, 2004). Individuals with DS that were diagnosed with OSAS based on PSG criterion, scored significantly lower on IQ tests (Fernandez & Edgin, 2013), thus suggesting a possible relationship between disrupted sleep due to OSAS and intellectual development.

Likewise, anecdotal evidence, as reported by parents of individuals with ASD, outline concerns about disturbed onset of sleep (insomnia), as well as daytime sleepiness. This problem may be arising for a number of reasons. Engelhardt et al. (2013), found an association of delayed sleep onset and media (video games, television, etc.) use towards bedtime. The researchers theorize that melatonin production is decreased as a result of bright backlight display screens from their televisions. In addition to these biological factors, such as lowered melatonin production and disrupted circadian rhythm cycles, poor sleep hygiene is attributed to disturbed sleep.

Given the atypical nature of sleep between both groups, observing individuals that have been diagnosed with both NDD’s may yield interesting findings. Parents of children with DS reported that they have observed autism-like behavior in their children. The relationship between these two NDD’s is not fully understood, nor heavily researched. Scientists are searching for more information regarding the comparisons and dissociations between ASD and DS. A frequency of 5-7% has been reported in dual diagnoses of this linkage, a predicted rate of every 1 in 20 individuals (Kent, Evans, Paul, Sharp, 1999; as seen in Capone, Grados, Kaufmann, Bernad-Ripoll, and Jewel, 2005).

It has been suggested that higher rates of ASD are prevalent in individuals with DS based on their learning disabilities (e.g., individuals with severe learning disabilities show a higher risk
for developing ASD mannerisms). Specifically, presence of ASD could potentially be a function of profoundly delayed intellectual development. However, the relationship between IQ and ASD has not been consistently reported. IQ has not been consistently found to be delayed in individuals with DS and ASD, making the hypothesis that lower IQ in DS can predict autistic tendencies unlikely. If all the children with both DS and ASD had IQs of below 20, the association might be explicable, but it would not explain why their disabilities were so profound (Howlin, Wing & Gould, 2008). People that demonstrate this have mild, moderate, and severe learning disabilities, as well as some form of Autism. Individuals with DS are predisposed, as a result of sleep disruption, to potentially have adverse effects on cognitive development (Breslin et al., 2014). Though there are not very many parallels that have already been established in the literature between sleep and cognition for both populations, hypotheses can be formed and tested based on the previous findings and developments.

Disrupted sleep has shown to create a variety of behavioral issues in individuals with DS and ASD. Understanding the implications of these variables within the groups could help improve quality of life through treatment and early intervention. By doing so, it may aid to alleviate the behavioral, cognitive, and health consequences that many individuals suffer from. Therefore, we aimed at understanding the relationship between ASD and DS, in regard to sleep and cognition.

The first aim of our study was to compare the mean sleep disturbance scores between the two groups, extracted from the CSHQ, to explore the nature of parent report on sleep. Secondly, we hypothesized that we would find a correlation between sleep disturbance scores, and verbal and nonverbal IQ in ASD and DS.
Methods

Participants

Participants were used as secondary analysis from a larger study in the Down syndrome Research Group Lab. There were two groups. Our focus group, individuals with Autism, and our comparison sample, individuals with Down syndrome.

Autism

To confirm diagnoses, individuals with ASD were administered the *Autism Diagnostic Observation Schedule-2* (ADOS-2; Rutter, DiLavore, Risi, Gotham, and Bishop, 2012) by a research-reliable experimenter. The ADOS-2 is a gold-standard diagnostic assessment that has four modules that measures social and communicative skills, as well as restricted and repetitive behaviors. The examiner determines which module should be administered based on the individual’s language and developmental level. The modules are categorized from subjects who are completely nonverbal, to children and adults who have fluent speech. The resulting scores were in the clinical range for all ASD subjects. Of the 27 individuals that participated in the primary study, exclusionary criterion was: not completing the CSHQ (n=5) and not living at home with parents (n=7). The remainder sample of ASD participants was (n=15).

Down syndrome

Participants with Down syndrome (n=40, ages 10-18) were medically diagnosed, as confirmed by karyotype. The total number of participants between both groups with exclusion factors considered is (n=36).

Materials
For this study, we used the Child Sleep Habits Questionnaire to determine eight specific domains of sleep, as well overall sleep disturbance. To measure intelligence, we administered the Kaufman Brief Intelligence Test-II. The scores of both measures were compared.

**Child Sleep Habits Questionnaire (CSHQ)**

This is a 33-item questionnaire that assess’ important aspects of sleep which may be disturbed. It has been used on a range of conditions from children to adolescents (Malow et al., 2009). The eight subscales are: “bedtime behavior and sleep onset; sleep duration; anxiety around sleep; behavior occurring during sleep; night waking; sleep-disordered breathing; parasomnias; and morning waking/daytime sleepiness” (Owens, Spirito, McGuinn, and Nobile, 2000). Total sleep disturbance score (TSDS) ranges from 33-103, with higher scores representing impaired sleep. The CSHQ was added at the end of the primary study. As a result, many parents answered the questionnaire over the phone, but some did fill it out independently, and sent it back. Parents interviewed over the phone were not prompted. They were read the direct question from the CSHQ. Parents answered to the best of their knowledge on their child’s sleep over the past week. The items are rated on a scale of “usually” (5-7 times per week), “sometimes” (2-4 times per week), and “rarely” (0-1 time per week). Higher scores on the CSHQ indicates higher rates of disturbed sleep. Some questions were reversed in order to rate the severity of sleep problems compared to non-disturbed sleep.

**Kaufman Brief Intelligence Test, Second Edition (KBIT-II)**

The KBIT-II is a neuropsychological examination that measures verbal intelligence (verbal knowledge and riddles) and non-verbal intelligence (matrices). Both verbal knowledge and riddles tests crystallized ability (knowledge of words and their meanings). Items cover both receptive and expressive vocabulary, and they do not require reading or spelling (Kaufman and
Kaufman, 1990). Matrices assess' fluid thinking by completing analogies and understanding relationships of topics (Kaufman and Kaufman, 1990). The mean standard score of the KBIT-II is 100, with standard deviation of 15. This test was administered in a laboratory setting by a research assistant, and takes roughly 20 minutes to complete. Standard scores are calculated for verbal and non-verbal IQ, as well as full-scale IQ. Higher scores on KBIT-II indicate a higher IQ for verbal and nonverbal standard scores.

**Statistical Analysis**

We used SPSS 20.0 to analyze the results of this study. Mean scores were calculated based on raw score for each of the 8 subscales on the CSHQ for the DS and ASD groups (see table 1). The clinical cut-off score was 41 on Total Sleep Disturbance Score (TSDS). ASD and DS had elevated scores on CSHQ; DS at 86% and ASD at 67% (see figure 1). A two-way ANOVA was conducted to analyze groups; comparing both, and within each individual group. We anticipated there would be within group significance for ASD based on the ADOS severity score. We ran nonparametric correlations of the CSHQ subscales compared with verbal and nonverbal IQ, as determined by KBIT-II scores (see table 2).

**Results**

We examined the relationship between mean scores for DS and ASD. We found ASD had significantly higher scores for daytime sleepiness $F (1, 34) = 6.04, p = 0.02$ and sleep onset delay $F (1, 34) = 4.40, p = 0.04$, compared to DS. We also found that DS was marginally significant in bedtime resistance $F (1, 34) = 4.06, p = 0.05$, compared to ASD (see figure 1). This finding corresponds with our hypothesis that sleep problems are severe for both groups. Results for ASD participants on daytime sleepiness, compared to DS, were interesting. "Children with ASD have problems with sleep onset, restless sleep, daytime sleepiness, with more stage shifts per hour on
PSG than children with Autism or typically developing (TD) children” (Richdale and Schreck 2009), which coincides with past research on sleep in Down syndrome. We found that daytime sleepiness in participants with ASD was significantly higher than in DS (p=0.02). In addition, sleep onset delay was higher in ASD than in DS (p=0.04). Insomnia is a highly reported parent-concern for Autism (Adams et al., 2014). We determined that at (p=0.05), bedtime resistance was marginally significant in favor of Down syndrome. No significance was found for sleep duration (p=0.40), sleep anxiety (p=0.23), night waking (p=0.33), parasomnias (p=0.91), and sleep disordered breathing (p=0.91).

We also wanted to determine if sleep differentially relates to cognitive function, particularly verbal IQ. Research has reported (small samples provided) that impaired cognitive functioning can be related to poor sleep in people with Autism (Richdale et al., 2009). We also examined the relationship between sleep subscales and IQ standard scores for verbal and nonverbal knowledge. There was marginal correlation between daytime sleepiness (p=0.45) and TSDS (p=0.49) correlated with verbal IQ for ASD (see table 2). We did not find any correlations between sleep and IQ in the DS population. When comparing correlation coefficients (cc) between groups, we did find a slight level trend between the two. ASD had a higher cc score compared to DS for sleep disordered breathing (ASD cc=0.38 compared to DS cc=-0.10) and TSDS (ASD cc=0.49 compared to DS cc=0.23) for verbal IQ. These trends help explore our hypothesis on the effects of sleep on cognitive function. The DS group showed higher cc for nonverbal IQ, compared to ASD, for sleep onset delay (DS cc=-0.21, ASD cc=0.03) and daytime sleepiness (DS cc=0.24 compared with ASD cc=0.09). Overall, there were no other significant differences or trending levels of sleep subscales compared to IQ in ASD and DS.

Discussion
General Discussion

The gaps within the literature still lie with understanding the relation between sleep and cognition in individuals with NDD. There has been extensive research completed on sleep for ASD and DS populations through use of: Sleep questionnaires, PSG studies, and sleep monitoring through activity counts (actiwatch). Williams, Sears, and Allard (2004) estimated a 44-83% prevalence of sleep disorders for ASD. To what extent behavior and cognition is affected by poor sleep, is still being considered.

This study examined two aims: the comparison of mean scores between Autism and Down syndrome based on parent report on the Child Sleep Habits Questionnaire, and the correlation between aspects of sleep (CSHQ subscales) and IQ (verbal and nonverbal) in both populations. The findings of our study can be concluded in three brief points. First, individuals with Autism have higher scores of daytime sleepiness and sleep onset when compared with Down syndrome. Second, a marginal correlation was found between verbal IQ and TSDS in Autism. Lastly, contrary to past, distinguished research, our results yielded a counter-intuitive correlation.

In our study, we first compared the two populations mean TSDS scores for the CSHQ. We found a significant difference for two subscales of sleep between ASD and DS, where ASD had higher scores for daytime sleepiness and sleep onset delay. These findings indicate that individuals with Autism have reportedly higher daytime sleepiness and sleep onset delay compared to DS. We also found that DS had higher scores for bedtime resistance. This finding indicates that these individuals with DS have higher reported scores of bedtime resistance. Carter et al. (2014) mentions individuals with DS showed “oppositional behaviour at bedtime and took an excessive time to settle to sleep” (2014). This coincides with the literature on ASD of high
prevalence of bedtime resistance. Among children with ASD, prolonged sleep latency and decreased sleep efficiency is the most common sleep problems (Adams et al., 2014). Poor sleep is known to be prevalent in both populations. However, our findings do not correctly represent past studies that show sleep disordered breathing being a prevalent factor for individuals with DS. Based on our findings, sleep deficits such as bedtime resistance and disrupted sleep is occurring in both populations. Although ASD shows a significant increase in severity of daytime sleepiness and sleep onset delay, the literature shows that individuals with DS are also having problems in these areas.

We ran a nonparametric correlation between verbal and nonverbal IQ, and sleep in both populations. The results showed a positive correlation (marginal) between verbal IQ and TSDS in the ASD group. This marginal correlation is indicating that the higher total sleep disturbance score (which indicates poor sleep), the higher the verbal IQ scores. This coincides with Richdale and Prior’s (1995) finding that children with ASD who had an IQ higher than 55, had greater sleep disturbances than those that had an IQ lower than 55. We did also find a slightly lower correlation between verbal IQ and daytime sleepiness in ASD. There were no other correlations found between IQ and sleep between both populations. However, when we compared correlation coefficients between groups, we did find that DS showed a higher correlation coefficient for two sleep subscales (sleep onset delay and daytime sleepiness) and nonverbal IQ. Likewise, ASD had higher correlation coefficients for sleep disordered breathing and TSDS in verbal IQ, when compared with DS.

**Critical Discussion**

It is compelling that we did not find any significant differences for mean sleep scores between groups on the other 5 subscales (i.e. sleep disordered breathing, parasomnias, night waking, sleep
anxiety, and sleep duration). Especially given how prevalent sleep disordered breathing is in DS. It is concerning that our parent reports show low levels of sleep disordered breathing in DS, as previous findings in the literature strongly support that individuals with DS suffer from OSAS. This indicates that both groups have high disturbed sleep and sleep anxiety. According to a literature overview by Adams et al. (2014), researchers are finding that these subscales are arising frequently in people with Autism. We think that the results from our findings (mean scores and correlation) touches upon the need to question the validity of parent report. Parent reported sleep is not a strong indication of disturbed sleep in their children because the disruption does not only happen at the onset of sleep, but can happen throughout the entire night. We question the validity of the CSHQ to effectively measure sleep in individuals with neurodevelopmental disorders. Owens et al. (2000) commented that one of the limitations of the CSHQ is that parent report is not always accurate at being aware of difficulties during sleep and sleep onset problems. More than half of the CSHQ’s were completed over the phone with the participant’s parent. Our study was a secondary analysis of an already completed study. Therefore, for time efficiency, the parents completed the questionnaire over the phone. To maintain accuracy, it would be beneficial to have the parents complete the questionnaire by hand. This may help ensure that the parent is not under-reporting on their child’s disturbed sleep because another person, which may make them feel “put on the spot”, is questioning them.

Future Directions

A future direction is to compare IQ in both populations with a more valid measurement of sleep, such as Polysomnography, which is a proven, reliable, objective means of monitoring the participant’s actual sleep. Given that parent report is inaccurate in correctly identifying disturbed sleep severity, all-night monitoring of the individual’s sleep will truthfully identify disturbed
sleep. Chen, Spano, Edgin (2013) stated that parent report did not correctly identify sleep habits in individuals with OSAS. This discrepancy confirms that parents are not great reporters on their child’s sleep behaviour. An additional direction we would like to see is an increase in sample size between both groups. ASD occurs more often in males than in females (Edwards, Watkins, Lotfizadeh, and Poling, 2012), which is representative of our sample. It would be beneficial to try to recruit more females to see the results of a larger, more diverse sample of ASD. There was variability between groups in regard to cognitive scores for ASD, as well as DS. A larger sample size would reduce the possibility of within-group differences, which could throw off the results of cognition and sleep.

Lastly, sleep interventions for individuals with NDD are essential. Early intervention for the person is critical for cognition. Direct effects of sleep on cognition for neurodevelopmental disorders are still being considered, but in our study and in the literature, there does seem to be an effect happening. For example, Andreou, Galanopoulou, Gourgoulianis, Karapetsas, Molyvdash (2002) found a relationship between the more apnoea’s a person (DS) has, the more difficulties they have in visuoperceptual skills. In opposition, Mayes & Calhoun (2009), reported that they discovered mixed findings in the literature on IQ and sleep in people with ASD. The literature continues to progress in understanding the weight that disrupted sleep holds on individuals with neurodevelopmental disorders. Given that quality of life is highly impacted for ASD and DS, in the realm of cognition and sleep, it is of benefit to the individuals, families, and scientists to research medical implications and early intervention strategies, so that these individuals can lead successful, healthy, and happy lives.
References


Chen, C., Spano, G., Edgin, J., (2013). The Impact of Sleep Disruption on Executive Function in Down syndrome. *Research in Developmental Disabilities*. 34 (e.g. 2), pp.6


Richdale, A., (2000). Sleep Problems in Autism: prevalence, cause, and intervention. *Developmental Medicine & Child Neurology. e.g. 32* (e.g. 2), pp.6


Figure Legends

Table 1.
The table shows the individual mean ratings for participants with ASD and DS on each scale of the CSHQ.

Figure 1.
The graph is displaying mean ratings between groups. The three subscales that had significance were: daytime sleepiness $F(1,34)= 6.04, p= 0.02$; sleep onset delay $F(1,34)= 4.40, p= 0.04$; and bedtime resistance $F(1,34)= 4.06, p= 0.05$.

Table 2.
The table is showing the correlation matrices for the ASD and DS. CSHQ sleep subscales are compared with standard score for verbal and nonverbal IQ, as determined by the *KBIT-II*. The tables’ show relationship (marginal significant correlation) between daytime sleepiness and total sleep disturbance score ($p= 0.06$) for verbal IQ in Autism. *Correlation is significant at the 0.05 level for two-tailed correlation.*
Table 1.

<table>
<thead>
<tr>
<th>Sleep Subscale</th>
<th>Down Syndrome Mean (n=21)</th>
<th>Autism Mean (n=15)</th>
<th>Total Mean (n=36)</th>
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<tr>
<td>Bedtime Resistance</td>
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<td>Sleep Onset Delay</td>
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<td>4.0278</td>
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<td>9.75</td>
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<tr>
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<td>4.3056</td>
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<td>Total Sleep Disturbance Score</td>
<td>49.0476</td>
<td>47.9333</td>
<td>48.5833</td>
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</tbody>
</table>

Figure 1.

Sleep Subscale Means Across Populations

- Child Sleep Questionnaire Scales
- * Down syndrome
- Autism

- Daytime Sleepiness
- Sleep Disordered Breathing
- Parasomnias
- Night Wakings
- Sleep Anxiety
- Sleep Duration
- Sleep Onset Delay
- Bedtime Resistance
Table 2.

<table>
<thead>
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<tr>
<td>CSHQ Bedtime Resistance Total</td>
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