Published in final edited form as:

J Intellect Disabil Res. 2017 March ; 61(3): 210–220. doi:10.1111/jir.12315.

Reliability of parent report measures of sleep in children with Down syndrome

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Abstract

Background—Behavioural sleep disturbances are common among children with Down syndrome (DS). However, tools used to detect and evaluate behavioural sleep disturbances were developed for typically developing children and have not been evaluated for use among children with DS. The current study evaluates the psychometric properties of three measures of behavioural sleep disturbances that are currently being used with children with intellectual and developmental disabilities, including children with DS.

Method—Caregivers of 30 children with DS rated their child's sleep with the Behavioral Evaluation of Disorders of Sleep (BEDS), Children's Sleep Habits Questionnaire (CSHQ) and Sleep Disturbances Scale for Children (SDSC). Caregivers also provided information on sleep diagnoses and completed a 7-night sleep and behaviour diary.

Results—The study investigated the rate of detecting sleep problems, internal consistency, and convergent and concurrent validity of the BEDS, CSHQ and SDSC. Children with DS were reported to exhibit behavioural sleep disturbances at different rates depending on the measure used; 0% BEDS, 79.3% CSHQ, and 17.2% SDSC. Internal consistency was comparable for all three measures for their total scores. However, when evaluating the internal consistency of subscale scores, those on the CSHQ and SDSC performed more strongly. The subscales of the CSHQ performed best when evaluating convergent and concurrent validity, with the SDSC subscales performing moderately well.

Conclusion—The study findings suggest that, among children with DS, the CSHQ and its subscales performed in a psychometrically sound and theoretically appropriate manner in relation to other measures of sleep, medical history of sleep problems, and daily reports of sleep and associated behaviours. The SDSC performed moderately well. When evaluating behavioural sleep disturbances among children with DS, the CSHQ is recommended based on its stronger psychometric properties.

Keywords

Down syndrome; trisomy 21; behavioural sleep disturbances; measurement; children

Children with Down syndrome (DS) are at risk for a variety of sleep problems. Obstructive sleep apnoea affects 31–66% of individuals with DS (de Miguel-Diez, Villa-Asensi, & Alvarez-Sala, 2003; Marcus, Keens, Bautista, von Pechman, & Ward, 1991; Maris, Verhulst, Wojciechowski, Van de Heyning, & Boudewyns, 2016; Stebbens, Dennis, Samuels, Croft, & Southall, 1991), and behavioural sleep disturbances, such as delayed sleep onset, night-time awakenings, and early morning awakenings, affect 52–69% of children with DS, and a potentially greater range (13–86%) of adults with DS (Carter, McCaughey, Annaz, & Hill, 2009; Churchill, Kieckhefer, Bjornson, & Herting, 2014; Epstein, Pillar, Tzichinsky, Here, & Lavie, 1992; Esbensen, 2016; Rosen, 2011; Stores & Stores, 1996). The rates of these sleep problems are higher than that observed in typically developing children, but still demonstrate significant variability between studies. The United States National Institutes of Health put together a working group to evaluate clinical outcome measures for use with individuals with DS. One of their observations was that measures of behavioural sleep problems warrant evaluation. Further, they identified the necessity of establishing the validity of measures of sleep for being able to detect treatment changes in clinical trials (NICHD, 2015). It is likely that the variety of measures used to detect behavioural sleep problems have contributed to the wide range in reported problems.

Obstructive sleep apnoea (OSA) is detected using polysomnography (PSG), and is the gold standard for both detecting OSA and for detecting improvements following surgical interventions to support OSA. In contrast, methods for detecting behavioural sleep disturbances in children with developmental disabilities currently report the frequency of sleep disturbances using a variety of different measures (Breslin, Edgin, Bootzin, Goodwin, & Nadel, 2011; Malow et al., 2006; Stores, Stores, Fellows, & Buckley, 1998). Parent report measures are most commonly used to assess behavioural sleep disturbances, with the preponderance of measures having been developed for typically developing children. Preliminary data suggest that parent reports are inadequate at assessing abnormal PSGs in children with DS, but these parent report measures have not been evaluated for detecting behavioural sleep disturbance in this population (Shott et al., 2006). Given the high and variable rates of sleep problems among children with DS and the impact sleep can have on daytime behaviours, there is a need to establish the psychometric properties of measures of behavioural sleep disturbances.

Children with DS are at greater risk of some behavioural sleep disturbances in contrast to adolescents or adults with DS. Younger children with DS experience more frequent night-time awakenings than adolescents (Carter et al., 2009). Although there is limited research examining sleep problems in relation to age in DS, there is a pattern of reports of lower rates of behavioural sleep disturbances in adults with DS than among children with DS (Esbensen, 2016). This higher rate of behavioural sleep disturbances in childhood highlights the need for a measure appropriate for use with this age range.

Several tools have been developed to detect behavioural sleep disturbances in typically developing children (Dorris, Scott, Zuberi, Gibson, & Espie, 2008; Spruyt & Gozal, 2011), yet only a few have been used with children with intellectual and developmental disabilities (IDD), including the Behavioral Evaluation of Disorders of Sleep (BEDS), the Children's Sleep Habits Questionnaire (CSHQ), and the Sleep Disturbances Scale for Children (SDSC)

(Bruni et al., 1996; Owens, Spirito, & McGuinn, 2000; Schreck, Mulick, & Rojahn, 2003). The BEDS has been used to describe sleep problems in children with autism spectrum disorders and Angelman syndrome (Polimeni, Richdale, & Francis, 2005; Schreck & Mulick, 2000; Taylor, Schreck, & Mulick, 2012; Walz, Beebe, & Byars, 2005). The CSHQ has been used to describe sleep problems in children with intellectual disabilities, autism spectrum disorders, Williams syndrome, and with DS (Ashworth, Hill, Karmiloff-Smith, & Dimitriou, 2013; Breslin et al., 2011; Breslin et al., 2014; Carter et al., 2009; MacCrosain & Byrne, 2009; Malow et al., 2006). The SDSC has been used among children with autism spectrum disorders, and also used to validate the Sleep Questionnaire developed by Simonds and Parraga (SQ-SP) among these children (Maas et al., 2011; Miano et al., 2007). While these measures are used among children with IDD, and the CSHQ is being used with children with DS, these measures have not been empirically validated for use among children with DS.

Given the high and variable rate of reported sleep problems and the need for sound measures of behavioural sleep disturbances for children with DS, this study aimed to evaluate the psychometric properties of three sleep measures, specifically the BEDS, CSHQ, and SDSC. First, we described the rates of different sleep problems detected by these measures among children with DS. Second, we examined the internal consistency of these three measures and their subscales. Third, we examined the convergent validity of the subscales in comparison to each other, and the overall measures in comparison to each other. Fourth, we examined the concurrent validity of the measures and their subscales in relation to clinical diagnoses of sleep apnoea, use of positive airway pressure (PAP), and 7-day sleep diary data. Understanding the psychometric properties of measures of behavioural sleep disturbances with children with DS will inform the use of reliable measures when reporting rates of sleep problems, and with detecting true change in sleep during behavioural interventions or clinical trials.

Method

Participants

Parents of 30 children with DS completed rating forms as part of a larger community-based study on sleep and associated daytime behaviour and cognition. Children with DS ranged in age from 6 to 17 years of age ($M = 11.68$ years, $SD = 2.73$), were primarily male (60%) and Caucasian (93.3%). Standard scores on the Kaufman Brief Intelligence Test-2 ranged from 40–65 ($M = 44.57$, $SD = 6.46$). Respondents were primarily mothers (96.6%).

Procedure

Families were recruited based on the age of the child and a diagnosis of Down syndrome. Parents provided information on the child's demographics and a brief medical history, including sleep diagnoses and sleep treatments. For seven consecutive nights at home, parents completed a sleep and behaviour diary regarding their child. The sleep and behaviour diary included parental report of how long it took the child to fall asleep (sleep onset latency), whether sleep behaviours were observed (night-time awakening, snoring, sleep apnoea episode, rhythmic jerking of legs), daytime drowsiness, and whether the child

had a nap the following day. Three measures of behavioural sleep disturbances were completed by parents at home, including the BEDS, CSHQ, and SDSC. The study was approved by the Institutional Review Board at the medical centre.

Measures

The Behavioral Evaluation of Disorders of Sleep (BEDS) is a 28-item measure of sleep problems for typically developing children that has been used with children with developmental disabilities (Schreck et al., 2003). Parents rate their child's sleep based on the last 6 months. Subscales include Expressive Sleep Disturbances, Sensitivity to the Environment, Disoriented Wakening, Sleep Facilitators, and Apnea & Bruxism. Internal consistency is adequate with typically developing children and children with autism spectrum disorders (Schreck & Mulick, 2000), and the factors discriminate between children with and without sleep disorders. Specifically, among typically developing children, the BEDS demonstrates good internal consistency for the total score ($\alpha = .82$) and poor to good for the subscales ($\alpha = .60-.85$). Items are rated on a 5-point scale from (0) never to (4) always.

The Children's Sleep Habits Questionnaire (CSHQ) is a 33-item sleep screening instrument for children and assesses major childhood medical and behavioural sleep disorders (Owens et al., 2000). The CSHQ provides ratings regarding Bedtime Resistance, Sleep Duration, Parasomnias, Sleep-Disordered Breathing, Night-Wakings, Daytime Sleepiness, Sleep Anxiety, and Sleep Onset Delay. Internal consistency is questionable to acceptable for the total score ($\alpha = .68$ with community, $\alpha = .78$ for clinic), unacceptable to acceptable for subscales with typically developing children ($\alpha = .36-.70$), and unacceptable to excellent for children with sleep disorders ($\alpha = .44-.93$). Test-retest over two weeks are poor to adequate for subscales ($r = .40-.79$). Items are rated on a 3-point scale from (1) rarely to (3) usually.

The Sleep Disturbances Scale for Children (SDSC) is a 26-item parent-completed rating scale of childhood sleep disorders (Bruni et al., 1996). Subscales include assessments of Disorders of Initiating and Maintaining Sleep, Sleep Breathing Disorders, Disorders of Arousal/Nightmares, Sleep/Wake Transition Disorders, Disorders of Excessive Somnolence, and Sleep Hyperhydrosis (excessive sweating). Among typically developing children, the SDSC is reported to have acceptable internal consistency for the total score, both with children with ($\alpha = .71$) and without ($\alpha = .79$) sleep disorders (Bruni et al., 1996). Test-retest reliability over approximately one month was also acceptable for the total score ($r = .71$). Correlations between subscales and the total score are unacceptable to acceptable ($r = .45-.77$) for children without sleep disorders, and lower for children with sleep disorders ($r = .31-.69$). The SDSC total score is reported to demonstrate concurrent validity with diagnosed sleep disorders, including insomnia, hypersomnia, respiratory disturbances during sleep, and parasomnias. The subscales demonstrate low inter-correlations. Items are rated on a 5-point scale from (1) never to (5) always. A cut-off of 39 (possible range 26–130) is proposed to have a good balance of sensitivity and specificity. The cut-off equates to a mean score of 1.5.

Data Analysis

For the purpose of the current analyses, mean scores were calculated for all measures and subscales. The clinical range of concern was defined as scores two standard deviations above the mean of the normative sample on which the BEDS, CSHQ, or SDSC were developed. As a total score of 39 is recommended as a cut-off score for the SDSC, this score was converted to a mean score and also used to evaluate children meeting clinical concern for sleep problems on the SDSC. Internal consistency was calculated using Cronbach's alpha.

Convergent validity was examined by correlating subscales on the BEDS, CSHQ, and SDSC that theoretically were related to each other. To assess *breathing concerns*, the following subscales were correlated: BEDS Apnea & Bruxism, CSHQ Sleep Disordered Breathing, and SDSC Sleep Breathing Disorders. To assess *sleep onset behaviours*, the following subscales were correlated: BEDS Sensitivity to Environment, CSHQ Bedtime Resistance, CSHQ Sleep Onset Delay, CSHQ Sleep Anxiety, and SDSC Initiating and Maintaining Sleep. To assess *arousal concerns*, the following subscales were correlated: BEDS Expressive Sleep Disturbances, CSHQ Night Waking, CSHQ Parasomnia, SDSC Disorders of Arousal, and SDSC Sleep-Wake Transition. And to assess *daytime drowsiness*, the following subscales were correlated: BEDS Disoriented Awakening, CSHQ Daytime Sleepiness, and SDSC Disorders of Somnolence. Convergent validity was also examined by correlating the total scores on the BEDS, CSHQ, and SDSC with each other.

Concurrent validity was examined by correlating subscales on the BEDS, CSHQ, and SDSC to clinical diagnoses of sleep apnoea and parent report of PAP. Concurrent validity was also examined by correlating subscales on the BEDS, CSHQ, and SDSC to individual items assessed on the sleep and behaviour diary, specifically how long it took the child to fall asleep, if the child woke frequently during the night, if the child snored loudly during the night, if the child stopped breathing during the night or woke up with a snorting sound, if the child demonstrated rhythmic jerking of their legs while they slept, if they appeared drowsy during the day, or if they took a nap.

Results

Frequency of Sleep Problems

Table 1 presents the mean subscale scores and the rates at which children met clinical criteria for individual subscales and the total scores on the BEDS, CSHQ, and SDSC.

On the BEDS total score, no children with DS were identified as having sleep behaviours in the clinical range. On the individual subscales, rates of sleep disturbances ranged from no children meeting clinical criteria for Expressive Sleep Disturbances, to 58.6% meeting criteria for Sleep Apnea & Bruxism.

On the CSHQ total score, 79.3% of children were identified as having behavioural sleep disturbances. On the individual subscales, rates of sleep disturbances ranged from 13.3% of children meeting clinical criteria for Sleep Onset Delay, to 33.3% meeting criteria for Sleep Anxiety.

On the SDSC total score, 17.2% of children were identified as having behavioural sleep disturbances (two standard deviations). In contrast, using the cut-off provided by the authors of the SDSC, 72.4% of children were identified as having behavioural sleep disturbances. On the individual subscales, rates of sleep disturbances ranged from 3.4% of children meeting clinical criteria for Sleep Hyperhydrosis, to 34.5% meeting criteria for Sleep-Wake Transition Disorders.

Internal Consistency

The alpha values for the total scores were questionable for the BEDS, and good for the CSHQ and SDSC (see Table 2). The alpha coefficients for the BEDS ranged from .331 (unacceptable) to .851 (good), for the CSHQ ranged from .599 (poor) to .864 (good), and for the SDSC ranged from .000 (unacceptable) to 1.000 (excellent). On the SDSC, the Disorders of Arousal scale had items with zero variance, contributing to the .000 internal consistency score. On the Sleep Hyperhydrosis scale of the SDSC, all parents rated the two items on this subscale identically.

Convergent Validity

Convergent validity between theoretically related subscales across measures of the BEDS, CSHQ, and SDSC provided variable results depending on the construct being evaluated. Significant correlations were found between specific subscale measures of *breathing concerns* and *daytime drowsiness* across the BEDS, CSHQ and SDSC (see Table 2). Regarding *sleep onset behaviours*, all specific subscales of the BEDS, CSHQ and SDSC demonstrated significant correlations, with the exception of the CSHQ Sleep Onset subscale. This subscale appears to measure the time delay to falling asleep in contrast to the other specific subscales which appear to assess sleep hygiene and associated behaviours that cause problems with falling asleep. In contrast, the specific subscales measuring *arousal concerns* on the BEDS, CSHQ, and SDSC generally did not demonstrate good convergent validity. The arousal subscale on the BEDS, CSHQ, and SDSC assess a variety of arousal behaviours, from being restless, to moving beds, to waking up screaming. The CSHQ Parasomnia subscale correlated with most other subscales assessing *arousal concerns* on the CSHQ and SDSC.

Convergent validity between total scores on the three sleep measures was consistent. The BEDS total score demonstrated significant correlations with the CSHQ total score, $r = .61$, $p = .001$, and SDSC total score, $r = .64$, $p = .000$, and the CSHQ total score demonstrated significant correlations with the SDSC total score, $r = .76$, $p = .000$.

Concurrent Validity

The presence of a sleep disorder, as identified by parents, was correlated with the CSHQ Sleep Disordered Breathing subscale and the SDSC Sleep Hyperhydrosis subscale but with no measures on the BEDS (see Table 3). Use of PAP was correlated with the CSHQ Sleep Disordered Breathing and Daytime Sleepiness subscales, with the SDSC Disorders of Somnolence and Sleep Hyperhydrosis subscales, but with no measures on the BEDS (see Table 3).

Variable correlations were found when examining the concurrent validity of the subscale scores in relation to individual sleep behaviours on the sleep and behaviour diary. On the BEDS, only the Sleep Apnea & Bruxism subscale was correlated with any sleep and behaviour diary ratings. Children rated high on this BEDS subscale also reported a higher rate of nightly snoring, stopping breathing, and motor jerking, and also being drowsy during the day (see Table 3).

On the CSHQ, several subscales demonstrated significant correlations with reports from the daily sleep and behaviour diaries. The Sleep Onset Delay subscale was correlated with nightly reports of how long it took the child to fall asleep and with daytime drowsiness. The CSHQ Sleep Duration subscale was correlated with nightly waking, stopping breathing, motor jerking, and daytime drowsiness. The CSHQ Night Waking and Parasomnia subscales were correlated with nightly waking. The CSHQ Sleep Disordered Breathing subscale was correlated with nightly snoring, stopping breathing, and motor jerking. The CSHQ Daytime Sleepiness subscale was correlated with daily reports of being drowsy during the day. The total score on the CSHQ was correlated with nightly waking, stopping breathing, motor jerking, and daytime drowsiness.

On the SDSC, several subscales also demonstrated significant correlations with reports from the daily sleep and behaviour diaries. The Disorders of Initiating and Maintaining Sleep subscale was correlated with nightly waking. The SDSC Sleep Breathing Disorders subscale was correlated with nightly snoring, stopping breathing, and motor jerking. The SDSC Disorders of Somnolence subscale was correlated with nightly waking, snoring, stopping breathing, and daytime drowsiness. The SDSC Sleep Hyperhydrosis subscale was correlated with nightly snoring, stopping breathing, and motor jerking. The total score on the SDSC was correlated with nightly waking, snoring, stopping breathing, jerking, and daytime drowsiness.

Discussion

The psychometric properties and validity of measures of behavioural sleep disturbances have received little attention among children with DS. Measures currently used were developed for typically developing children and demonstrate poor ability to detect sleep apnoea in children with DS. We evaluated three measures of sleep currently being used with children with IDD or DS specifically, including the BEDS, CSHQ, and SDSC, for their ability to detect behavioural sleep disturbances in an effort to guide the use of appropriate measures among children with DS.

When comparing the three sleep measures on internal consistency, detection of sleep problems, and validity, the CSHQ and SDSC appear to demonstrate the stronger psychometric properties when measuring behavioural sleep disturbances in children with DS. Internal consistency was comparable for all three measures for their total scores. However, when evaluating the internal consistency of subscale scores, those on the CSHQ and SDSC performed stronger.

The CSHQ also appeared to be most consistent with rates reported in the literature of behavioural sleep disturbances. The ability of the subscales to detect severe symptoms two standard deviations above the mean of normative data varied among the BEDS, CSHQ, and SDSC. The BEDS total score detected no children with severe behavioural sleep disturbances, whereas on the BEDS subscales, over half of the children were reported to score high on the Sleep Apnea & Bruxism subscale. The low rate of detecting severe behavioural sleep disturbances in children with DS is inconsistent with rates reported in the literature (Breslin et al., 2011; Carter et al., 2009). At the other extreme, the CSHQ total score detected over three-quarters of children demonstrating overall behavioural sleep disturbances. On the subscales of the CSHQ, the rates of behavioural sleep disturbances ranged from 13–33%, which is a finding more consistent with the rates of concerns noted in the literature on children with DS (Breslin et al., 2011; Carter et al., 2009). The rate at which the total score on the SDSC identified children with severe behavioural sleep disturbances fell between the rates reported on the BEDS and CSHQ, identifying just fewer than 20% of children. When using the recommended cut-off of 39 on the SDSC, a comparable rate (72.4%) of overall behavioural sleep disturbances in children with DS was identified as on the CSHQ. In addition, some of the SDSC subscales identified behavioural sleep disturbances at the same rate as the CSHQ subscales, specifically the SDSC subscales of Sleep Breathing Disorders, Disorders of Initiating and Maintaining Sleep, and Sleep-Wake Transition Disorders. The lower rates on the other subscales were not unexpected. Excessive sweating (hyperhydrosis), night-time awakening with screaming, and sleepwalking are not often reported in the literature for children with DS. However, the low rate of Disorders of Excessive Somnolence on the SDSC was unexpected given the reported concerns in the literature on children with DS for daytime drowsiness (Carter et al., 2009). When screening for sleep problems in children with DS, scores two standard deviations above the mean on the CSHQ or using the total score cut-off of 39 on the SDSC are recommended.

The CSHQ subscales performed the best when assessing the validity of the behavioural sleep measures, with the SDSC also performing moderately well. All three measures demonstrated strong convergent validity for their total scores with each other. Further, the subscales of the three measures also generally demonstrated strong convergent validity, particularly on the subdomains of sleep disordered breathing, sleep onset, and daytime drowsiness. However, for concurrent validity, the CSHQ demonstrated the stronger psychometric properties in comparison to the BEDS and SDSC. The CSHQ subscales correlated as expected with medical history of sleep problems and with measures from daily sleep and behaviour diaries. In contrast, the SDSC subscales did not correlate with medical history of sleep problems, and not all correlations with daily sleep and behaviour diaries made logical sense. Similarly, the BEDS did not correlate with medical history of sleep problems, or with most measures from daily sleep and behaviour diaries.

Specifically, when examining the domain of *sleep disordered breathing*, the BEDS Apnea & Bruxism, CSHQ Sleep Disordered Breathing, and SDSC Sleep Breathing Disorders demonstrated good convergent validity with each other. Further, all three subscales demonstrated good concurrent validity with daily sleep diary reports of the child stopping breathing, snoring, and motor jerking during sleep. However, only the CSHQ Sleep Disordered Breathing was related with a medical history report of a current diagnosis of a

sleep disorder and current use of PAP, demonstrating the strength of the subscales of the CSHQ in contrast to those on the BEDS and SDSC in assessing sleep disordered breathing.

For *daytime drowsiness*, the BEDS Disoriented Awakening, CSHQ Daytime Sleepiness, and SDSC Disorders of Excessive Somnolence were all significantly inter-correlated, demonstrating good convergent validity. With regard to concurrent validity, both the CSHQ Daytime Sleepiness and SDSC Disorders of Excessive Somnolence, but not the BEDS Disoriented Awakening, were significantly correlated with daily behaviour diaries of daytime drowsiness. While, the SDSC Disorders of Excessive Somnolence was significantly correlated with daytime drowsiness, it also demonstrated significant correlations with night-time awakening, snoring, and stopping breathing. While these additional correlations are not unexpected, they do suggest the subscale does not discriminate as effectively in assessing daytime behaviours. Thus, the CSHQ subscales demonstrate the strongest psychometric properties in detecting daytime drowsiness.

For *sleep onset*, again the subtests of the BEDS, CSHQ, and SDSC demonstrated good convergent validity. The BEDS Sensitivity to Environment, CSHQ Bedtime Resistance, CSHQ Sleep Anxiety, and SDSC Disorders of Initiating and Maintaining Sleep were all significantly inter-correlated. These subscales all measure behaviours associated with sleep hygiene or difficulties with settling to sleep, and were not measured on the daily sleep diaries. The CSHQ Sleep Onset Delay subscale did not correlate with these other subscales of sleep onset, as it does not measure the behaviours associated with difficulties falling asleep, but rather the duration it takes a child to fall asleep. When assessing concurrent validity, the CSHQ Sleep Onset Delay was the only subscale to significantly correlate with daily sleep diary reports of the latency to fall asleep, supporting that this subscale assesses sleep latency and not behavioural difficulties with sleep initiation. The SDSC Disorders of Initiating and Maintaining Sleep was significantly correlated with the daily sleep diary report of waking during the night, suggesting that this subscale accurately detects night-time awakening in addition to difficulties with sleep onset. Thus, both the subscales on the CSHQ and SDSC demonstrate sound psychometric properties in detecting difficulties with sleep onset in children with DS.

For *arousal*, the subscales on the BEDS, CSHQ, and SDSC performed variably for convergent and concurrent validity. The BEDS Expressive Sleep Disturbances subscale was not correlated with the other measures of arousal on the CSHQ and SDSC. The two CSHQ subscales (Night Waking and Parasomnias) were inter-correlated, as were the two SDSC subscales (Disorders of Arousal and Sleep Wake Transition Disorders), suggesting that subscales were more strongly correlated within a measure (CSHQ or SDSC) than within the construct (arousal). Only the CSHQ Parasomnia subscale demonstrated good convergent validity with subscales of other measures, specifically both the SDSC subscales (Disorders of Arousal and Sleep Wake Transition Disorders). Further, only the CSHQ subscales (Night Waking and Parasomnias) demonstrated good concurrent validity with the daily sleep diary reports of night-time awakening, while the BEDS and SDSC subscales assessing arousal behaviours did not correlate with daily sleep diary reports. Again, the subscales of the CSHQ demonstrated better psychometric properties in detecting sleep arousal in children with DS.

Some subscales on the CSHQ and SDSC did not fit within these theorised domains, but were still evaluated for concurrent validity with daily sleep diaries. The CSHQ Sleep Duration subscale was significantly correlated with nightly waking, stopping breathing, motor jerking, and daytime drowsiness. As this subscale measures children receiving too little sleep, these significant correlations were theoretically appropriate. The SDSC Sleep Hyperhydrosis subscale, measuring excessive sweating, was also significantly correlated with nightly waking, stopping breathing, and motor jerking.

The number of individuals with DS used to evaluate the psychometric properties of the BEDS, CSHQ, and SDSC was small. Even with the small sample size, there was evidence for strong psychometric properties for the CSHQ and strong to modest properties for the SDSC. Further, ratings of sleep from multiple parents were not available. Future research warrants an evaluation of the inter-rater reliability of these behavioural measures of sleep. Despite these limitations, there are numerous strengths to the current study. Our study is the first psychometric evaluation of measures used to detect behavioural sleep disturbances among children with DS, and to evaluate the validity of both total and subscale scores of these measures. Demonstrating preliminary evidence of strong psychometric properties of the CSHQ validates its use in current studies of children with DS (Carter et al., 2009), and has implications for ensuring the accurate detection of behavioural sleep disturbances in children with DS. Being able to support the use of measures of behavioural sleep disturbances also ensures a more accurate evaluation of treatment effects. Thus, our findings have implications for clinical trials and for behavioural therapy targeting to improve sleep outcomes for children with DS.

This study contributes to our understanding of measurement of sleep in children with DS, a topic of importance for accurately assessing symptoms and evaluating outcomes for children with DS (NICHD, 2015). Our preliminary findings indicate that the CSHQ and its subscales performed in a theoretically appropriate manner in relation to other measures of sleep, medical history of sleep problems, and daily reports of sleep and associated behaviours. The SDSC performed modestly, and is to be recommended for screening for overall sleep problems using the total score cut-off. In contrast the BEDS did not currently demonstrate consistent reliability and validity to recommend its use in children with DS. However, the BEDS may be appropriate for assessing sleep in children with other IDD, and warrant additional psychometric evaluation with these populations of children. These measures all warrant further evaluation in other populations of individuals with IDD. While selecting one measure to assess sleep problem may be ideal for making comparisons of sleep problems across different genetic syndromes, another measure may be better at assessing outcomes from clinical intervention to support sleep problems within a specific syndrome group. Currently, when evaluating behavioural sleep disturbances among children with DS, the CSHQ is recommended and demonstrates strong psychometric properties within this population.

Acknowledgments

This manuscript was prepared with support from the *Eunice Kennedy Shriver* National Institute of Child Health and Human Development of the National Institutes of Health (R21 HD082307, A. Esbensen, PI), the Jack H Rubinstein Foundation, and the Emily Ann Hayes Research Fund. The content is solely the responsibility of the authors and

does not necessarily represent the official views of the National Institutes of Health. This research would not have been possible without the contributions of the participating families.

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Table 1

Mean scores, frequency of meeting clinical criteria, and internal consistency on the BEDS, CSHQ and SDSC.

	Mean (SD)	Range	Met clinical criteria	Alpha
BEDS (range 0–4)				
Expressive sleep disturbances	.11 (.15)	0–0.5	0%	.352
Sensitivity to the environment	.58 (.53)	0–1.88	6.9%	.549
Disoriented awakening	1.27 (1.00)	0–3.75	6.9%	.851
Sleep apnea & bruxism	.87 (.81)	0–2.67	58.6%	.331
Total	.54 (.32)	0–1.04	0%	.695
CSHQ (range 1–3)				
Bedtime resistance	1.44 (.52)	1–2.83	26.7%	.802
Sleep Onset Delay	1.43 (.73)	1–3	13.3%	^a
Sleep duration	1.42 (.60)	1–3	23.3%	.864
Sleep anxiety	1.51 (.56)	1–2.75	33.3%	.654
Night Waking	1.61 (.66)	1–3	31%	.793
Parasomnia	1.35 (.29)	1–2	27.6%	.599
Sleep Disordered Breathing	1.35 (.47)	1–3	31%	.713
Daytime Sleepiness	1.65 (.38)	1–2.5	21.4%	.661
Total	1.48 (.25)	1.06–2.03	79.3%	.803
SDSC (range 1–5)				
Disorders of initiating & maintaining sleep	1.89 (.52)	1–2.86	20.7%	.529
Sleep breathing disorders	1.71 (.73)	1–3.67	20.7%	.766
Disorders of arousal	1.10 (.24)	1–2	6.9%	.000 ^b
Sleep wake transition disorders	1.81 (.66)	1–3.17	34.5%	.711
Disorders of somnolence	1.72 (.55)	1–3.2	6.9%	.607
Sleep hyperhydrosis	1.24 (.79)	1–5	3.4%	1.000 ^c
Total	1.68 (.36)	1.04–2.46	17.2% [*]	.800

^aSubscale contains only one item

^bZero variance in parental responses to two items

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c, Parent responses identical

* Using the recommended cut-off score for the SDSC (total score 39, mean score 1.5), 72.4% of children met clinical criteria for behavioural sleep disturbances.

Table 2

Correlations between subscales of the BEDS, CSHQ, and SDSC.

	1	2	3	4	5	6	7	8	9	10	11	12
Breathing												
1 BEDS Sleep apnea & bruxism												
2 CSHQ Sleep Disordered Breathing	.60**											
- SDSC Sleep breathing disorders	.69**	.84**										
Sleep Onset												
3 BEDS Sensitivity to the environment												
4 CSHQ Bedtime resistance			.60**									
5 CSHQ Sleep Onset Delay			.02	.03								
6 CSHQ Sleep anxiety			.65**	.73**	-.20							
- SDSC Disorders of initiating & maintaining sleep			.46*	.41*	.36	.45*						
Arousal												
7 BEDS Expressive sleep disturbances								.20				
8 CSHQ Night Waking							.20	.57**				
9 CSHQ Parasomnia							.30	.25	.38*			
10 SDSC Disorders of arousal							.28	.33	.84**	.39*		
- SDSC Sleep wake transition disorders												
Daytime Drowsiness												
11 BEDS Disoriented awakening											.44**	
12 CSHQ Daytime Sleepiness											.59**	.64**
- SDSC Disorders of somnolence												

* $p < .05$,

** $p < .01$

Table 3

Significant correlations between BEDS, CSHQ, and SDSC with sleep medical history and sleep/behaviour daily diary.

	Medical History		Sleep & Behaviour Diary						
	Sleep Disorder	PAP	Time to sleep	Woke	Snore	Stopped breathing	Jerked	Drowsy in day	Nap in day
BEDS									
Expressive sleep disturbances									
Sensitivity to the environment									
Disoriented awakening									
Sleep apnea & bruxism					.521 **	.458 *	.638 **	.436 *	
Total									
CSHQ									
Bedtime resistance									
Sleep Onset Delay			.658 **					.487 **	
Sleep duration				.492 **		.402 *	.566 **	.401 *	
Sleep anxiety									
Night Waking				.575 **					
Parasomnia				.430 *					
Sleep Disordered Breathing	.434 *	.420 *			.755 **	.693 **	.603 **		
Daytime Sleepiness		.380 *						.468 *	
Total				.533 **		.375 *	.397 *	.431 *	
SDSC									
Disorders of initiating/maintaining sleep				.414 *					
Sleep breathing disorders					.667 **	.530 **	.595 **		
Disorders of arousal									
Sleep wake transition disorders				.448 *	.404 *	.416 *		.419 *	
Disorders of somnolence									
Sleep hyperhydrosis		.430 *			.845 *	.721 *	.744 *		
Total	.400 *	.652 **		.508 **	.456 *	.549 **	.540 **	.476 **	

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*,⁵⁰ > *p*
*,⁵⁰ > *p*
*,⁵⁰ > *p*

PAP = positive airway pressure; Time to sleep = how long it took the child to fall asleep; Woke = child woke frequently during the night; Snore = child snored loudly during the night; Stopped Breathing = child stopped breathing during the night or woke up with a snoring sound; Jerked = child demonstrated rhythmic jerking of their legs while they slept; Drowsy = child appeared drowsy during the day; Nap in day = child took a nap.