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Modified Children's Sleep Habits Questionnaire for Behavioral Sleep Problems: A Validation Study

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Abstract

Objectives—Behavioral sleep problems (BSPs) are prevalent and consequential in young children. There is a need for screening tools that identify BSPs- which are often rooted in the parent-young child relationship- and typically respond to behavior management. Such a tool would increase capacity to identify and treat BSPs. We sought to validate a short-form version of the widely used Children's Sleep Habits Questionnaire (SF-CSHQ) that omitted items that would not be responsive to behavioral strategies.

Methods—The original 33-item CSHQ elicits parent report of "behaviorally-based" and "medically-based" sleep items (e.g., parasomnias and sleep disordered breathing). We conducted analyses to develop a SF-CSHQ that excludes its "medically-based" items, to determine a) the SF-CSHQ threshold score corresponding to the full CSHQ clinical cut-off score (41), and b) preliminary validity of this SF-CSHQ. Data were re-analyzed from the original data that established the CSHQ's psychometric properties in 4-10 year olds, and a second dataset that established its validity in 24-66 month olds.

Results—In both datasets, a threshold score of 30 had correlations of 0.90-0.94 with the original cut-off. This 23-item SF-CSHQ cut-off functioned as well as the full CSHQ cut-off in discriminating between children with vs. without a parent-reported behavioral sleep problem, and with vs. without prolonged sleep latency (per actigraphy).

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Conclusion—We established preliminary validity of modified version of the widely used CSHQ. This SF-CSHQ may be useful for widening screening and first-line guidance for behavioral sleep problems in young children, among professionals who are not sleep medicine specialists.

Introduction

Insufficient and poor quality sleep in young children adversely impact behavioral and cognitive function. Preschool age (3-5 years) is a peak time for sleep disturbances, when $\approx 25\%$ of children have a behavioral sleep problem (BSP).(1, 2) BSPs are problems falling or staying asleep, such as bedtime resistance, night-waking; they correspond to the "insomnia" diagnosis under the 2014 classification of sleep disorders.(3) (Previously, "sleep onset association" and "limit setting" types of behavioral insomnia were classified separately.(4)) Among children with neurodevelopmental disorders, prevalence is even higher, up to 80%. (5, 6) BSPs impede development of executive function,(7) and may increase later need for special education. However, healthy sleep practices (e.g. regular bedtime, bedtime routine), and behavioral interventions (e.g., extinction) both promote healthy sleep (8) and address sleep problems.(9) In fact, most BSPs in young children do not require specialized attention (10) Other sleep problems in young children--- sleep-disordered breathing (SDB) and parasomnias-- are not amenable to brief behavioral interventions. Parasomnias often resolve spontaneously with age, (11) while alternative gold-standard screening tools exist for SDB (12-14), which is managed by weight loss, medications, and surgery. (15)

Given the prevalence of BSPs in early childhood and their responsiveness to behavioral interventions, there is a need for criterion-referenced screening tools (10). Secondary prevention for mild BSPs could be accomplished by training a range of health professionals to deliver brief, behavioral interventions.(16) Positive effects are found when such interventions are delivered to parents by nurses and psychology trainees, (9, 17) via the internet,(18-20) and via written materials.(21) Behavioral interventions are most effective in younger (vs. older) children,(21) perhaps because they target parent-child interactions that contribute to the problem.(22)

The CSHQ is one of the most widely used multi-dimensional tools used to screen for pediatric sleep problems. It was designed to reflect common clinical symptoms presenting in school aged (4-10 year) children. Using a clinimetrics approach, i.e., one based upon symptoms or clinical presentation (23), the CSHQ includes 33 distinct items grouped into 8 subscales, based on face and content validity: bedtime resistance, sleep-onset delay, sleep duration, sleep anxiety, night-waking, parasomnias, SDB, and daytime sleepiness. Parents indicate the frequency of the sleep behavior during a typical week as 'Usually' [5-7 times/ week], 'Sometimes' [2-4 times/week], or 'Rarely' [0-1 times/week]. Higher scores indicate worse sleep behaviors or problems. The CSHQ's validity was originally evaluated for two groups of 4-10 year olds: a community sample of elementary school children (n=469), and a sleep disorders clinic sample of 154 children, including those with a BSP (n=43), parasomnia (n=45), or SDB (polysomnography [PSG] confirmed, n=66). A total score 41 discriminated the two samples with acceptable sensitivity (0.80) and specificity (0.72).(24) Further details are presented in Methods.

The CHSQ is used in diverse community, general pediatric, and condition-specific populations. It was developed as a research tool; it has been employed in more than 300 research projects. It has been translated into at least 19 languages with adequate to good reliability and/or validity in Chinese,(25), Dutch (26), Portugese (27) and Spanish. (28) Notably, a recent paper employed the Spanish version as the gold standard for determining the validity of the shorter BEARS screener, in primary care practice.(29)

The current study aims to provide preliminary validity for a shortened version of the CSHQ (SF-CSHQ), exclusive of its parasomnia and SDB items. Such a tool would be specific to BSPs, thus identifying common sleep problems that are responsive to behavioral interventions delivered to (and administered by) parents. The 23-item SF-CSHQ evaluated in this study excludes its parasomnia (n=7) and SDB (n=3) items. A modified CSHQ excluding these items has been used by others,(30) and detected significant change after a BSP intervention.(30, 31) However, researchers who employed this modified CSHQ did not establish validity of a clinical cut-off for it. Here, we aim to determine a) the SF-CSHQ threshold score corresponding to the full CSHQ clinical cut-off score (41), and b) preliminary validity of this SF-CSHQ.

Methods

We re-analyzed datasets from two studies that had utilized the CSHQ. The first (Owens) was the original study of community and sleep disorder clinic sample data, collected in 1997-1998, that validated the CSHQ in 4-10 year olds. Analyses presented here utilize data from the sleep disorder clinic sample. (24) The second dataset collected in 2003-2005 (Goodlin-Jones)(32) included, in addition to the full CSHQ, actigraphy, sleep log, and a single-item sleep measure for younger children, aged 24-66 months. Authors of both studies agreed to share these de-identified datasets for analysis.

Description of Datasets

Owens—Total score internal consistency (α) for the community (.68) and sleep clinic (.78) samples was high using the criterion of 0.70.(33) For the bedtime resistance, sleep duration, sleep anxiety and daytime sleepiness scales, internal consistency was comparable in the community (0.63-0.70) and clinic (0.68-0.80) samples. The SDB subscale performed poorer in the community (0.51) vs. clinic (0.93) sample as did the parasomnia subscale (community=0.36, clinic= 0.56). In contrast, the night-waking subscale performed better in the community (0.54) vs. the clinic (0.44) samples. Sleep onset delay (> 20 minutes) was assessed from a single-item, thus no alpha was computed. Supporting the CSHQ's validity, the sleep clinic sample had higher (worse) scores on the total and all subscale scores, controlling for age and SES. Sensitivity and specificity were maximized using a cut-off score of 41 under the Receiver Operator Characteristic (ROC) curve.(24)

The CSHQ's development and psychometric properties have been examined by others. Of 57 pediatric sleep questionnaires analyzed, the CSHQ fulfilled 5 of 11 criteria for tool development; just two questionnaires fulfilled all steps.(12) Further, psychometric properties vary by population. In Dutch school age children, CSHQ internal consistencies ranged from 0.47-0.68, though test-retest and inter-observer reliabilities were good.(26) Among 2-5 year

olds, a third of whom included early intervention or mental health program participants, subscale alphas ranged from 0.55-0.82, with, the parasomnia (0.69) and night-waking (0.68) subscales performing better than in the original community sample.(34)

Goodlin-Jones—We analyzed data from a study used to establish the full CSHQ's validity in young typically and non-typically developing children. (32) Families were recruited to a study about "sleep and waking patterns" (vs. a study of sleep problems) from community settings and a neuro-developmental disorders research registry.(35) Study participants reported on here include children who are typically developing (n=73), with autism (n=83), or with developmental delay without autism (n=64). The sample's full CSHQ total score had high internal consistency, 0.82. Additional sleep data included: actigraphy, a sleep log, and a single-item global assessment. Children wore actigraphs for 7 days and nights, from which the following sleep variables were derived: sleep start time, total 24 hr sleep duration, nightwaking duration, and night wake-up times. Actigraphy data were averaged over one week of observations. Parents recorded sleep start and wake-up times as well as night-waking and nap durations in sleep logs on days their child wore the actigraph. Finally, parents were asked "Does your child have a sleep problem at the current time? (Yes/No)." The CSHQ significantly correlated with actigraphy-derived sleep onset (0.48), night-waking duration (0.21), total 24 h sleep (0.25), awake time (0.62), and; discriminated parents reporting their child's sleep as problematic (Yes vs. No).(32)

Statistical Analyses

To determine the SF-CSHQ threshold score that corresponds to the full CSHQ clinical cutoff score (41) we calculated linear regressions of the reduced scores on the full scores in both datasets. We calculated the threshold for the SF-CSHQ by using the regressionpredicted value when the full CSHQ equals 41. We report agreement between the percentage of positive screens obtained by the full and SF-CSHQ in both datasets, using the Cohen's Kappa (standard error [SE]).

Next, we examined the SF-CSHQ's ability to discriminate children with vs. without a parentreported sleep problem per a single-item global measure (Yes vs. No), in Goodlin-Jones' data. Such single-item measures are associated with sleep problems (i.e., onset, duration, night-waking),(36) sleep logs, the CSHQ,(37) and may correlate more highly with a child's mood and functioning than objective measures (e.g., PSG, actigraphy). (37) On a population basis, they predict quality of life, learning and behavioral outcomes at school-entry.(38)

We calculated the global measure's sensitivity and specificity to discriminate the two groups of children, using the area under the Receiver Operating Characteristics (ROC) curve. Sensitivity and specificity refers to a test's ability to detect "true positives" and "true negatives," respectively. The area under the ROC curve describes the ability of a continuous measure to discriminate a dichotomous outcome. When the continuous measure is expected to be higher among those with the target outcome, the value ranges from 0.5 -1.0, with 0.5 indicating no systematic prediction, and 1.0 indicating the existence of a perfect cutoff, above which all subjects have the targeted outcome, and below which none do. In practice,

0.70-0.79 is considered fair discrimination, 0.80-0.89 is good, and 0.90-1.0 is excellent.(39) Measures < 0.70 are not considered useful predictors in most contexts.

Finally, we conducted additional analyses to establish preliminary validity. In the Goodlin-Jones data, we calculated Pearson correlation coefficients between the full CSHQ and SF-CSHQ scores, and actigraph measures of sleep efficiency, night-wakings, awake time, and sleep latency. We also used Pearson coefficients to determine associations between a newly derived dichotomous SF-CSHQ measure, and these actigraphy-derived measures. Using the Owens data, we sought to determine whether the SF-CSHQ could identify sleep problems independent of SDB in the subset of children (n=66) with PSG-confirmed SDB. We analyzed correlations between hourly pauses in respiration (apnea-hypopnea index [AHI]) and the SBB items of the CSHQ.

Results

Descriptions of the two samples are shown in Table 1. Both were majority male (Owens, 70.9%; Goodlin-Jones 74.8%). Mean ages in the Owens (sleep clinic) and Goodlin-Jones samples were 8.0 and 3.7 years, respectively. The percentage of children scoring above the SF-CSHQ cut-off was 81.1% and 67.2% in the Owens and Goodlin-Jones samples, respectively. Agreement between the percentage of cases falling above the cut-off, using the original and short form measures was higher in the Goodlin-Jones (0.80, SE= 0.07) compared to the Owens (0.41, SE= 0.08) sample. Given the predominance of SDB and parasomnia diagnoses in the Owens data (72%), this supports the utility of the SF-CSHQ as a measure of BSPs.

In the Goodlin-Jones data, we found that E(reduced score)= -4.12 + 0.83*full score, R² = 0.94. Accordingly, a threshold value of 41 for the full score corresponds to a reduced scale score of approximately 30. In the Owens data, we found that E(reduced score)= -6.1 + 0.80* (full score), R² = 0.89. The somewhat lower strength of association may reflect the treatment of questionnaire item non-response. In the Owens data, missing item responses were imputed when < 20% of items were missing. In contrast, in the Goodlin-Jones data, only participants providing complete data on all items were included in the scoring.

By developmental status, the full CSHQ (cutoff= 41) and SF-CSHQ (cutoff= 30) yielded comparable rates of sleep problems in the Goodlin-Jones data. These full CSHQ vs. SF-CSHQ rates were, respectively, 70 % vs. 72% for children with autism, 74% vs. 68% for children with developmental delay, and 59% vs. 62% for typically developing children. Overall, SF-CSHQ scores were higher in the Owens sleep clinic (Mean= 36.0, SD= 6.8) versus Goodlin-Jones community and neuro-developmental sample (Mean= 33.7, SD= 7.3). There were no differences in SF-CSHQ scores by gender. [Data not shown.]

Using the Goodlin-Jones "Yes vs. No" global measure as the "gold standard" we calculated the sensitivity, specificity and area under the ROC curve for both CSHQ versions (Table 2). Using a threshold value of 41 for the full-scale score, the CSHQ has sensitivity 90.8% and specificity 45.5%. Using a threshold value of 30, the SF-CSHQ has sensitivity 89.2% and specificity 44.6%. For the full-scale score, the area under the ROC curve was 0.80 (95% CI=

0.74-0.87). For the SF-CSHQ, it was 0.79 (95% CI= 0.73-0.86) for the SF-CSHQ. Boxwhisker plots of the two scale distributions in each group of children are shown in Figure 1.

Associations between actigraphy measures (Goodline-Jones data) and the CSHQ are shown in Table 3. The correlation of the continuous SF-CSHQ score with sleep latency was statistically significant: r=0.26, p<.05. [For the continuous full-scale score, correlation to sleep latency was r=0.24 (p<.05)]. Correlations with other actigraph measures were small and did not reach significance. Sleep latency was longer among children whose SF-CSHQ score exceeded the newly derived cut-off (41 min, SD= 30), than those whose scores did not (34.8 min, SD= 18.8). As with linear measures, the dichotomous measure of the SF-CSHQ did not discriminate children by the number of night-wakings, awake time, or sleep efficiency. Use of the full-scale dichotomized CSHQ (41) produced comparable results for these actigraphy-derived measures.

In the Owens sample, correlations between the SF-CSHQ, sleep medications, and sleep variables derived from the full CSHQ (total sleep time, sleep efficiency, and night-waking duration) were low, as shown in Table 4. Note, this sample consists primarily of children with a parasomnia or PSG-confirmed SDB (72% vs. 28% with a BSP). In the subsample of children with PSG-confirmed SDB, we confirmed that the SF-CSHQ can identify sleep problems independent of SDB, because it has very low correlations with both the SDB related subscale of the CSHQ [r=-0.01, p=0.90] and the apnea-hypopnea index [r=-0.09, p=0.19] of the PSG. By contrast the SDB items of the CSHQ omitted from the SF-CSHQ correlate moderately strongly with the AHI, r= 0.29, p=0.02 (not shown). Additionally, in this Owens subsample of children with PSG-confirmed SDB, the correlations between SF-CSHQ scores and sleep behaviors (i.e., sleep efficiency, wake duration) were low and not significant. These results favor convergent discriminant validity of the SF-CSHQ as a pure measure of sleep disturbance (not shown).

Discussion

Criterion-referenced tools are needed to identify children with mild BSPs for whom brief interventions are often effective. The CSHQ is the most commonly used tool to screen for multi-dimensional sleep problems in children. All but its parasomnia and SDB domains reflect areas of concern that are primarily rooted in parent-child interactions, and are responsive to behavioral interventions. We re-analyzed data from two datasets used to validate the CSHQ- initially and in younger children-to determine a short form CSHQ (SF-CSHQ) cut-off score excluding parasomnia and SDB items, and to establish its preliminary validity as a screener for parent-reported BSPs. In both datasets, a score of 30 had correlations of 0.90-0.94, based on 23 of the original 33 items. The SF-CSHQ cut-off score functioned as well as the full CSHQ cut-off in discriminating between children with vs. without a parent-reported sleep problem, and between children with vs. without prolonged sleep latency as measured by actigraphy. (12, 26, 34) Furthermore, there was higher agreement between the two versions re: the percentage of positive screens in the Goodlin-Jones sample, compared with the full Owens sample which was predominately children with diagnosed SDB or parasomnia.

The SF-CSHQ shared the limited ability of the full CSHQ to predict actigraph outcomes, similar to studies comparing parent reports with objective measures.(40, 41) High correlations are found between actigraphy and sleep logs for sleep onset and wake-times (42-44); correlations for night-waking are less robust.(41, 45) Regarding PSG, studies find significant associations with the CSHQ (40) while others do not. (41) Objective measures emphasize sleep quality. Subjective reports reflect what is impactful for parents,(46) and what may respond to sleep hygiene education. CSHQ insomnia subscale scores improve after parent education, as do actigraphy measures.(31) Even for SDB, parental assessments were better predictors of SDB resolution than either PSG, clinical, or lab measures.(47) Thus, subjective assessments have a role distinct from that of objective measures.

This study has a number of strengths. We obtained and analyzed original data from two datasets that are key to validation of the CSHQ, originally and in younger children. Both included an objective assessment of sleep (actigraphy in one, PSG in the other) that we utilized in analysis. Furthermore, both datasets were utilized both to determine the SF-CSHQ threshold score, and to validate its use. The study also has limitations. Data for both were collected at least a decade earlier. Pediatric sleep medicine has seen changes in respiratory scoring of PSG,(48) and continued lack of standardized scoring for actigraphy. (49) Also, differences between the datasets in regarding measures (e.g., developmental diagnoses in one but not the other) and analysis (e.g., one suppressed an entire subscale's data if any one of its items was missing, the other did not) precluded conducting the exact same analyses in both. Conversely, such triangulation of data sources and methods, tends to strengthen the validity and applicability of findings.

One in four young children experience insomnia-related sleep difficulties. Most are transient and respond to brief educational interventions to improve sleep hygiene. Practically speaking, the SF-CSHQ is inexpensive, quick to administer, does not require computer scoring or unique credentialing and thus may be a viable screening option in wide variety of settings. Next steps might include further refinement of the SF-CSHQ such as: validation using more recent data, identification of the most discriminative items, and examination of its clinical utility. Such work would lend confidence to adding the SF-CSHQ to the toolkit of resources non-sleep specialists could use to widen secondary prevention efforts.

Even with further validation of the SF-CSHQ, there remains few evidence-based protocols (vs. information or advice) that non-sleep specialists can use to manage common sleep problems once identified. (10) A recent workshop highlighted the gap between knowledge in sleep science, and implementation of preventive interventions. One recommendation was to bolster educational interventions aimed at promoting awareness of the adverse health, functional outcomes and costs of sleep problems.(50) An ongoing study to promote sleep health literacy throughout early childhood programs seeks to bridge this gap. (51) Further work like this is needed to bridge the knowledge/action gap among school-aged children and adolescents.(52) Our findings support use of the SF-CSHQ to screen for behavioral sleep problems in settings where appropriate referral and/or delivery of evidence-based protocols by appropriately trained non-sleep specialists is available.

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Figure 1. Distributions of Full and SF-CSHQ Scores by Parental Report (Yes/No) of Sleep Problem

	Table 1
Demographics and	Short-Form CSHQ Scores

	Owens (Clinic Sample) (n=151)	Goodlin-Jones (N=218)
Female, n (%)	44 (29.1%)	55 (25.2%)
Male, n (%)	107 (70.9%)	163 (74.8%)
Age, Mean (sd)	8.0 years (3.1)	3.7 years (0.93)*
SF-CSHQ, Mean (SD)	36.0 (6.8)	33.7 (7.3)
SF-CSHQ, percentage above cut-off	81.1%	67.2%
Agreement (Cohen's Kappa), percentage above cutoff sleep using full CSHQ and SF-CSHQ versions	0.41 (SE, 0.08)	0.80 (SE, 0.07)

* Goodlin-Jones dataset contains date of birth, but not date of participation. Thus, age can not be calculated. Age data shown are for the n=194 children reported on in Goodlin-Jones, 2008.

Table 2	
Validity Measures of Full and SF-CSHQ vs. Parental Report (Yes/No) of Sleep I	Problem

	Goodlin-Jones (N-218)
	Goodini-Jones (11-210)
Full CSHQ	
Sensitivity	90.8%
Specificity	45.5%
Area under ROC	0.80 (95%CI, 0.74-0.87)
Short Form (SF) CSHQ	
Sensitivity	89.2%
Specificity	44.6%
Area under ROC	0.79 (95% CI, 0.73-0.86)

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Table 3

Actigraphy Associations with Full and Short Form (SF) CSHQ, by Linear and Dichotomous CSHQ Measures

	Lin	ear				
Goodlin-Jones Data	Full CSHQ (linear) (Pearson r)	SF-CSHQ (linear) (Pearson r)	Full CSHQ 41 Mean (SD)	Full-CHSQ < 41 Mean (SD)	SF-CSHQ 30 Mean (SD)	SF-CSHQ < 30 Mean (SD)
Sleep Efficiency: Percent of time in bed that person is awake	-0.13	-0.12	89.0 (6.2)	89.8 (6.8)	89.2 (6.3)	89.5 (6.7)
Night-Waking: Duration in minutes	-0.04	-0.08	2.9 (1.8)	3.3 (2.3)	2.9 (1.8)	3.4 (2.3)
Awake Time: Per 24 hour clock	0.07	0.04	21.4 (8.0)	20.2 (17.6)	21.0 (18.0)	20.9 (17.7)
Sleep Latency: Minutes to fall asleep	0.24 *	0.26 *	41.9 (29.7)	33.5 (18.1)	41 min (30)*	34.8 min (18.8) *

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F-CSHQ, Sleep Medication and Sleep Measure

	SF-CSHQ	Sleep Medications	Total Sleep Time	Sleep Efficiency	Wake Duration
SF-CSHQ	1.0000				
Sleep Medications	$0.04 \ P = .81 \ N = 39$	1.0000			
Total Sleep Time	$^{-0.49}_{P=0.70}$ N = 103	-0.03 P = 0.87 N = 40	1.0000		
Sleep Efficiency	$\begin{array}{c} 0.09\\ P=0.36\\ N=99 \end{array}$	-0.02 P = 0.90 N = 39	0.47 P < 0.005 N = 144	1.0000	
Wake Duration	$^{-0.10}_{ m P} = 0.34$ N = 100	0.05 P < 0.76 N = 37	-0.33 P < 0.005 N = 143	-0.46 P < 0.005 N = 139	1.0000