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Original Article

# Behavioral sleep disturbances in children clinically referred for evaluation of obstructive sleep apnea

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ABSTRACT

*Objective/Background:* Obstructive sleep apnea (OSA) and behavioral sleep disturbances (BSD) are known to have a negative health impact on children. OSA and BSD may coexist; however, such comorbidity is not fully appreciated in clinical settings.
*Methods:* Patients referred for OSA evaluation completed polysomnography and the Children's Sleep Habits Questionnaire. Prevalence estimates for clinically significant BSD were computed and comorbidity of BSD and OSA was examined. Chart reviews were completed to determine if BSD were addressed in the medical treatment plan.
*Results:* Over one-half of the sample had a clinically significant BSD. Patients with comorbid OSA and BSD represented 39.46% of the sample. In 36–54% of the patients with a clinically significant BSD, no plan to treat the BSD was documented in the patient's medical record.
*Conclusions:* Children referred for evaluation of OSA have a high likelihood of experiencing clinically significant BSD irrespective of OSA diagnosis. Sleep medicine clinicians should be careful not to overlook the potential impact of BSD even after a child has been formally diagnosed with OSA. Physician knowledge of empirically supported behavioral sleep treatments or access to behavioral sleep medicine services is an essential component of comprehensive care for children clinically referred for OSA evaluation.


## 1. Introduction

Increased awareness regarding the impact of sleep disturbances on the health status of children has been achieved in large part due to research focused on pediatric obstructive sleep apnea (OSA). OSA is a respiratory sleep disorder characterized by airway obstruction that disrupts ventilation during sleep. The hallmark symptom of OSA is habitual (nightly) snoring. OSA affects 1–4% of children [1] and is recognized as a likely cause of significant medical morbidity (e.g., cardiac hypertrophy [2], inflammation [3,4], metabolic syndrome [5]) and changes in neuropsychological and behavioral functioning in children [6–10]. Nonrespiratory sleep disturbances as a group have higher prevalence rates than OSA. The most common nonrespiratory sleep disorders include parasomnias (e.g., sleep walking; sleep terrors; nightmares), sleep related movement disorders (e.g., periodic limb movement disorder; restless leg syndrome), circadian rhythm disorders (e.g., delayed sleep phase syndrome), disorders of excessive daytime sleepiness (e.g., narcolepsy), and behavioral sleep disturbances (e.g., behavioral insomnia of childhood). Behavioral sleep disturbances (BSD) in children rank highest among problems brought to the attention of pediatricians [16]. The most common BSD which include bedtime resistance, problematic sleep associations, and prolonged nocturnal awakenings have estimated prevalence rates ranging from 25% to 40% of children and adolescents [11–16]. Unresolved BSD may persist and develop into a chronic sleep disorder [17–19]. Similar to pediatric OSA, children with BSD have been observed to be at increased risk for neurobehavioral problems such as impairments in cognition, mood, attention, and behavior [20–22].

Historically, the presence of BSD in children with OSA was considered inconsequential [23]. Furthermore, the consensus clinical practice guideline for the diagnosis and management of childhood OSA [24] does not emphasize the importance of thoroughly screening for comorbid BSD when completing an OSA diagnostic evaluation. The primary emphasis of an OSA evaluation is on respiratory symptoms and airway obstruction. Use of overnight polysomnography (PSG) in a sleep laboratory has been considered the gold standard for diagnosing OSA [24]. Thus, in clinical practice the detection and treatment of BSD may be underemphasized when children are referred for evaluation of OSA.

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Several recent studies provide converging evidence that comorbid OSA and BSD are common in pediatric clinical samples [21,25–28]. Furthermore, it has been observed that children with comorbid OSA and BSD may be at greater risk for clinical morbidity than children with either OSA or BSD alone [26]. For example, Owens et al. [21] examined sleep and daytime behavioral functioning in school age children with OSA or a BSD who were clinically referred for sleep evaluation. In their clinical sample, comorbid OSA and BSD were documented in 22% of the patients formally diagnosed with OSA; externalizing behavior problems were more severe among children with comorbid OSA and BSD compared to children with OSA alone [21]. Given the potential medical and neurobehavioral sequelae associated with OSA and BSD, a better understanding of the comorbidity of these problems has clinical relevance but is not well documented. Owens et al. have published the most salient clinical research examining sleep comorbidities in children with OSA. In a recent study with a clinical sample of children who completed PSG for evaluation of OSA, it was found that the greatest predictor of behavioral morbidity in the sample was the presence of at least one comorbid sleep diagnosis in addition to OSA. Comorbid sleep disorders included movement disorders, parasomnias, hypersomnia, insomnia, and circadian rhythm disorders [27]. We identified only one previous study that has specifically examined the comorbidity of BSD and OSA in a sample of children clinically referred for sleep evaluation [21]. These findings support a recognized need to conduct further research in this area [29].

In the current study we sampled patients that were referred for OSA evaluation in the absence of other known sleep complaints because we suspected that the risk of overlooking BSD in this group was higher than children presenting with general sleep complaints. We also employed a clinical cut-off score for identifying patients with clinically significant BSD, as opposed to relying solely on mean score differences on paper–pencil measures. In addition, we were interested in determining the extent to which sleep physicians addressed BSD within the context of an OSA evaluation in light of anecdotal observations that BSD are not routinely detected and treated in children evaluated for OSA. The overarching goals of this study were to better understand the comorbidity of BSD and OSA in children clinically referred for OSA evaluation and to examine whether or not BSD were identified and treated during routine clinical care. The specific aims of the study were to (1) document parent-reported sleep problems in children clinically referred for OSA evaluation, (2) characterize the prevalence of clinically significant BSD in the clinical sample, (3) quantify the number of patients diagnosed with comorbid OSA and BSD, and (4) determine the extent to which BSD were recognized and addressed in the medical treatment plan for these children.

## 2. Methods

### 2.1. Participants

The sample was comprised of children aged 4–12 years referred for evaluation of OSA at an accredited sleep disorders center during 36 consecutive months from 2004–2006. Data included in this study were obtained via retrospective chart review that was approved by the institutional review board at the institution. The sleep disorders center is housed in a pediatric tertiary care hospital located in the mid-west and is staffed by pediatric pulmonologists board certified in sleep medicine and a pediatric psychologist certified in behavioral sleep medicine. The sleep disorders center provides clinical evaluation and management services for the broad spectrum of pediatric sleep disorders including both respiratory and non-respiratory conditions. All referrals were reviewed by the sleep disorders center medical director and were triaged based on referral concern/question and relevant symptoms of sleep disturbance. In addition to considering information provided by the referring physician, triage decisions were also guided by historical information and by caregiver reported concerns documented during a brief telephone interview. The phone interview between the child's primary caregiver and the sleep center administrative coordinator documents caregiver concerns regarding the child's sleep including specific symptoms of sleep-disordered breathing, sleep initiation, sleep maintenance, and sleep sufficiency.

In our clinical practice patients with symptoms of OSA who are referred for sleep apnea evaluation and do not have known symptoms of behavioral sleep problems at the time of referral are triaged for evaluation with a pulmonary sleep physician. Patients with symptoms suggestive of both OSA and BSD (e.g., bedtime resistance, negative sleep associations, trouble falling asleep, anxiety at bedtime, prolonged/problematic night wakings) are triaged for evaluation with a pulmonary sleep physician and the sleep disorders center psychologist. For the purposes of this research protocol, children known to have behavioral sleep complaints at the time of referral were excluded from the study. This sampling strategy was chosen in order to reduce the likelihood that patients included in the study would have a BSD based on clinical information known at the time of referral. Inclusion criteria were as follows: (1) child had no known behavioral sleep complaints at the time of referral, (2) child underwent clinical evaluation by a board certified pulmonary physician, and (3) child completed an overnight polysomnography. There were no explicit exclusion criteria.

During the 36 month data collection period, a total of 308 patients were identified as eligible based on the inclusion criteria. During this same time period 231 patients were referred for concerns that included obstructive sleep apnea, nonrespiratory sleep disorders, and behavioral sleep disturbances as opposed to obstructive sleep apnea in the absence of any other sleep concerns; these subjects were not included in the retrospective chart review because they did not meet study inclusion criteria. Of the total eligible patients ($n = 308$), 161 children were not included in the final data analysis due to missing or incomplete data. The final study sample consisted of 147 children who completed a comprehensive evaluation (see Section 2.2) for obstructive sleep apnea. The study sample represented 47.7% of the total eligible patients. There were no statistically significant differences between the final sample ($n = 147$) and the patients not included due to missing or incomplete data ($n = 161$) with respect to obstructive apnea–hypopnea index, age, and gender; AHI ($t[306] = -.09$; P = NS); Age ($t[306] = 1.03$; P = NS), and gender ($X^2 = .745$; P = NS). Demographic and clinical characteristics of the sample are summarized in Table 1.

### 2.2. Procedures

All patients included in the study were initially seen for clinical evaluation by a board certified pulmonologist and were subsequently referred for PSG to rule out obstructive sleep apnea. At the time of initial evaluation, the primary caregiver for each patient filled out our center's history form, the Sleep Disorders Center History Questionnaire (SDCHQ), and the Children's Sleep Habits Questionnaire (CSHQ), a validated pediatric sleep questionnaire. During the initial evaluation the SDCHQ and CSHQ were reviewed by the sleep physician and used to facilitate documentation of the clinical history for each patient. It is noteworthy that the CSHQ was not scored during the context of routine clinical care and thus clinicians may have reviewed responses to the CSHQ but did not consider CSHQ subscale or total scores when making a clinical diagnosis. The CSHQ was formally scored as part of the current research protocol and was used to document parent-reported sleep problems and to identify patients with clinically significant BSD.

**Table 1**
Demographic and clinical characteristics of the sample.

| Total sample ($N$ = 147) | | | |
|---|---|---|---|
| Age: mean (SD), range | 8.4 years (±2.5), 4–12.6 | | |
| Gender problems | | Family history of sleep | |
| Male | 62.6% | Sleep apnea | 71.4% |
| Female | 37.4% | Excessive sleepiness | 15.0% |
| Race | | Restless sleep | 17.7% |
| Caucasian | 70.7% | Parasomnia | 21.1% |
| African–American | 19.7% | Insomnia | 23.8% |
| Other | 9.5% | | |
| Medical history | | | |
| Asthma | 34.0% | | |
| Developmental delay | 33.3% | | |
| Seizures | 22.4% | | |
| Acid reflux | 24.5% | | |

Abbreviations: SD, standard deviation.

### 2.3. Clinical measures

#### 2.3.1. Children's Sleep Habits Questionnaire (CSHQ)

The CSHQ [30] is a 33 item paper–pencil parent report instrument that examines sleep behavior and symptoms of sleep disorders in children. This measure was developed for use in school-aged children ranging in age from 4 to 10 years [30]. Other published studies have demonstrated its use in younger (2–5.5 years) and older (10–16.9 years) children [31,32]. The CSHQ was developed considering the most prevalent pediatric sleep disorder diagnoses from the International Classification of Sleep Disorders [33]. The CSHQ includes items relating to the sleep domains that encompass the major sleep related concerns in children: bedtime behavior and sleep onset, sleep duration, sleep related anxiety, behaviors during sleep, night wakings, sleep disordered breathing, parasomnias, and daytime sleepiness. Caregivers use a 3-point Likert scale to answer questions. The measure yields a total score and 8 subscales scores that reflect key sleep domains that are representative of the primary medical and behavioral sleep disorders in children. Higher scores represent more problematic sleep. Psychometric properties published on this instrument demonstrate adequate internal consistency (.68–.78) and test–retest reliability within an acceptable range (.62–.79). Alpha coefficients ranged from .36 to .93 for the various subscales. Individual items, subscale items, and the total score have been shown to differentiate clinical from control groups demonstrating validity of the measure. Subscale reference values for clinical and community control samples were published along with the psychometric properties of the instrument [30]. A total score >41 on the CSHQ has been identified as the most sensitive clinical cut-off for identifying sleep problems in children [30].

The CSHQ was used to document parent-reported sleep problems and to measure clinically significant behavioral sleep disturbances (BSD) in the sample. In the validation study of the CSHQ, children with behavioral sleep disorders had significantly higher scores on 4 of the 8 CSHQ subscales (1. Bedtime resistance; 2. Sleep onset delay; 3. Sleep duration; 4. Sleep anxiety), relative to clinically referred children diagnosed with parasomnias or sleep-disordered breathing [30]. These four subscales have been demonstrated to be most characteristic of behaviorally based sleep problems [30] and thus were used to create cut-off scores for identifying clinically significant BSD in this study (see data analytic plan for further explanation). Hereafter these four subscales are referred to as the Children's Sleep Habits Questionnaire – Behavioral Sleep Disturbances (CSHQ–BSD) subscales.

#### 2.3.2. Sleep Disorders Center History Questionnaire (SDCHQ)

The SDCHQ was developed by the clinical staff at our center and was used to obtain relevant information pertaining to the patient's personal and family medical history and demographic information. Information is documented by the primary caregiver using open ended questions, multiple choice format for demographic information (age, gender, and race), and yes/no (presence/absence) responses regarding any history of relevant medical conditions (asthma, developmental delay, seizures, acid reflux) or family medical history of sleep problems (sleep apnea, excessive sleepiness, restless sleep, parasomnia, and insomnia). The SDCHQ was used for identifying the demographic and clinical characteristics of the sample.

#### 2.3.3. Lab based overnight polysomnography (PSG)

PSG studies were performed during a single overnight stay in the sleep disorders center sleep laboratory. PSG studies were completed according to the American Thoracic Society standards [34,35] using computerized systems. The formal interpretation of studies was completed by a board certified sleep physician. A diagnosis of OSA was assigned using diagnostic criteria for an obstructive apnea–hypopnea index (AHI) greater than one event per hour of sleep.

### 2.4. Data analytic plan

#### 2.4.1. Data pre-processing

SPSS PASW Statistics software, version 17 was used for all data analysis. Data pre-processing included computing total and subscale scores for the CSHQ. On the CSHQ, missing data were typically the result of the omission of one or two items on a multi-item subscale. In cases where less than 50% of subscale items were missing, composites were prorated from the remaining items for the subscale [32]. If more than 50% of the item level responses making up a subscale were missing, all data for that case were eliminated from final data analysis. Statistical comparisons were made between the final sample ($N$ = 147) and patients excluded from final data analysis due to incomplete CSHQ data ($n$ = 16). Age ($t$[161] = −.722; P = NS), gender ($X^2$ = .964; P = NS), and race ($X^2$ = 1.072; P = NS) composition of the two groups were not statistically significant.

#### 2.4.2. Covariate analyses

Spearman's correlation coefficients were calculated to test for an association between the obstructive apnea–hypopnea index (AHI) and CSHQ–BSD subscales to verify that these variables did not covary and represented distinct sleep disturbances. All sociodemographic and clinical variables (see Table 1) were evaluated as potential control variables (covariates). Spearman's correlation coefficients (between continuous variables) or point-biserial correlation coefficients (between continuous and dichotomous variables) were calculated to test for associations between all sociodemographic and clinical variables and CSHQ–BSD subscales. In all subsequent data analyses covariates were controlled for when required. All statistical tests were evaluated at the $p < .05$ significance level.

#### 2.4.3. Parent-reported sleep problems (Aim 1)

Descriptive statistics for all CSHQ subscale scores and the CSHQ total score were examined. $T$-tests were used to compare mean subscale scores for the entire sample to published community control reference values [30] (Table 2). The mean CSHQ total score was compared to the published clinical cut-off score of 41 that is considered a sensitive clinical cut-off for identification of probable sleep problems [30] (Table 2).

#### 2.4.4. Prevalence of clinically significant BSD (Aim 2)

The study sample was subdivided into patients with and without polysomnographically confirmed OSA. ANOVA was used to

compare mean scores on the CSHQ–BSD subscales between children with and without OSA (Table 3).

Owens et al. [30] paper describing the psychometric properties of the CSHQ included a community control sample that was younger than our study sample (mean age 7.6 versus 8.4 years) and had a gender distribution with less male subjects (51.2% versus 62.6% of the sample). Despite these differences, Owens' community control sample is the most comparable sample that we could identify in the published literature on the CSHQ. The CSHQ does not have published norms. We calculated clinical cut-off scores using the mean and standard deviation scores from CSHQ subscales. In accordance with the standard convention for many behavioral measures, we defined clinically significant behavioral sleep disturbances as CSHQ–BSD subscale scores >2 standard deviations above the published community control reference mean values [30].

Prevalence rates were computed for clinically significant BSD using the CSHQ–BSD subscales (Table 4). Chi-square analyses were performed to examine the distribution of prevalence rates for clinically significant BSD in children with and without OSA (Table 4).

*2.4.5. Comorbidity of clinically significant BSD and OSA (Aim 3)*

Using an AHI index >1 event per hour of sleep to diagnose OSA and a clinical cut-off score for defining the presence of a clinically significant BSD (CSHQ–BSD subscale score >2 SD above the community control reference value for the subscale) the sample was classified into one of four diagnostic subgroups: (1) OSA only, (2) Comorbid OSA/BSD, (3) BSD only, (4) No OSA/BSD. Descriptive statistics for each of the subgroups were examined (Fig. 1).

*2.4.6. Treatment of clinically significant BSD (Aim 4)*

Retrospective medical chart review was completed for each patient. Clinic notes related to each patient's initial evaluation and follow-up clinical care were coded to indicate whether or not behavioral sleep problems were addressed by the physician directly or through referral to the sleep center psychologist. Any reference in the physician's documentation to sleep hygiene recommendations, sleep schedule changes, stimulus control therapy, limit setting, behavioral modification, or referral to psychology were coded positively for BSD treatment (i.e., BSD was addressed in management plan). Descriptive statistics are reported for patients determined to have a clinically significant BSD (Table 5).

## 3. Results

### 3.1. Covariate analyses

There were no statistically significant correlations between the CSHQ–BSD subscales and AHI (range of $r = -1.160$ to $-0.45$; P = NS), indicating that these measures represented distinct measures of sleep disruption. There were no significant associations between child gender or race and any of the CSHQ–BSD subscales (range of $r = -.108$–$.77$; P = NS). Child age was associated with the CSHQ – Bedtime Resistance Subscale ($r = -.350$; $P = .000$) and the CSHQ – Sleep Anxiety Subscale ($r = -.305$; $P = .000$), indicating older children had lower levels of bedtime resistance and sleep related anxiety. With respect to family and medical history, the CHSQ – Sleep Onset Delay subscale was associated with family history of excessive daytime sleepiness ($r = -.187$; $P = .023$) and family history of insomnia ($r = -.246$; $P = .003$); the CSHQ – Sleep Duration Subscale was associated with family history of insomnia ($r = -.199$; $P = .016$) and child's medical history of gastroesophageal reflux ($r = -.189$; $P = .028$); the CSHQ – Sleep Anxiety Subscale was associated with child's medical history of asthma ($r = -.171$; $P = .049$); although $r$ values were small (<.3 in all cases), each of these statistically significant correlations indicated that the presence of the designated family and medical history variables were associated with higher scores (i.e., more problems) on the specific CSHQ–BSD subscales. No other statistically significant correlations were found at the $p < .05$ significance level.

**Table 3**
CSHQ Behavioral Sleep Disturbance subscales for children with and without OSA.

| Subscale score | OSA group | | Non-OSA group | | Univariate, *F* | *P* value |
|---|---|---|---|---|---|---|
| | Mean ($n = 87$) | SD | Mean ($n = 60$) | SD | | |
| Bedtime resistance | 9.01 | 2.75 | 9.34 | 2.79 | .48 | NS |
| Sleep onset delay | 1.67 | .74 | 1.88 | .761 | 2.97 | NS |
| Sleep duration | 5.61 | 1.97 | 5.62 | 1.77 | 0.00 | NS |
| Sleep anxiety | 6.28 | 2.36 | 7.00 | 2.46 | 3.22 | NS |

Abbreviations: CSHQ, Children's Sleep Habits Questionnaire; SD, standard deviation; OSA, obstructive sleep apnea, NS, not statistically significant.

**Table 4**
Prevalence rates for clinically significant[a] CSHQ–BSD subscale scores ($N = 147$).

| CSHQ subscale | Prevalence rate (%) | Pearson chi-square[b] | *P* value |
|---|---|---|---|
| Behavioral sleep problems | | | |
| Bedtime resistance (n = 43) | 29.3 | 1.62 | NS |
| Sleep onset delay (n = 28) | 19.1 | 1.21 | NS |
| Sleep duration (n = 76) | 51.7 | 0.00 | NS |
| Sleep anxiety (n = 47) | 31.9 | 1.03 | NS |

Abbreviations: CSHQ, Children's Sleep Habits Questionnaire; BSD, Behavioral Sleep Disturbances; NS, not statistically significant.

[a] Cases with CSHQ subscale scores >2 SD above control mean [30].

[b] Chi-Square analyses compared distribution of prevalence rates between OSA and non-OSA groups *n* = number of cases with CSHQ subscale scores >2 SD above community control reference values [30].

**Table 2**
CSHQ scores compared to normative data.

| CSHQ Score ($N = 469$) | Our sample ($N = 147$) | | Owens [30] control sample | | | |
|---|---|---|---|---|---|---|
| | Mean | SD | Mean | SD | 95% Conf. Interv. | |
| | | | | | Lower | Upper |
| Bedtime resistance subscale | 9.15* | 2.76 | 7.06 | 1.89 | 1.64 | 2.54 |
| Sleep onset delay subscale | 1.76* | .76 | 1.25 | .53 | .38 | .63 |
| Sleep duration subscale | 5.61* | 1.88 | 3.41 | .93 | 1.89 | 2.51 |
| Sleep anxiety subscale | 6.57* | 2.42 | 4.89 | 1.45 | 1.29 | 2.08 |
| Night wakings subscale | 5.30* | 1.86 | 3.51 | .89 | 1.49 | 2.09 |
| Parasomnias subscale | 11.11* | 2.26 | 8.11 | 1.25 | 2.63 | 3.37 |
| Sleep-disordered breathing subscale | 6.37* | 1.97 | 3.24 | .63 | 2.81 | 3.46 |
| Daytime sleepiness subscale | 16.33* | 3.68 | 9.64 | 2.80 | 5.99 | 7.23 |
| Total score | 58.95** | 9.55 | – | – | 16.3 | 19.45 |

Abbreviations: CSHQ, Children's Sleep Habits Questionnaire; SD, standard deviation.

* *T*-test comparing sample means was significant, $p < .001$.

** Total score for control sample not reported; sample mean score > clinical cut-off score of 41.

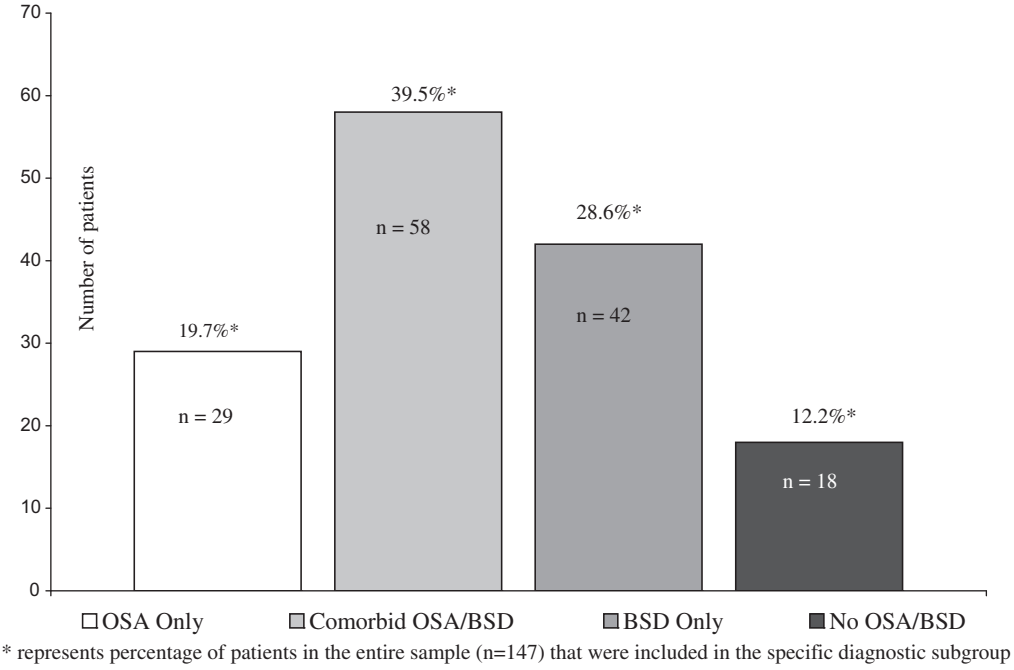


**Fig. 1.** Diagnostic categories. ∗Represents percentage of patients in the entire sample ($n$ = 147) that were included in the specific diagnostic subgroup.

**Table 5**
Clinically significant BSD cases with documentation of behavioral treatment.

| CSHQ subscale | $n$ | M.D.[a] | Ψ Referral[a] | M.D. and/or Ψ[a] |
|---|---|---|---|---|
| Bedtime resistance | 43 | 19 (44.2%) | 18 (41.9%) | 23 (53.5%) |
| Sleep onset delay | 28 | 8 (28.6%) | 10 (35.7%) | 10 (35.7%) |
| Sleep duration | 76 | 25 (32.9%) | 23 (30.3%) | 31 (40.8%) |
| Sleep anxiety | 47 | 16 (34.0%) | 13 (27.7%) | 19 (40.4%) |

Abbreviations: BSD, Behavioral Sleep Disturbances; Ψ, referral to sleep psychologist.
$n$ = number of cases with CSHQ subscale scores >2 SD above normal reference values [30].
[a] Number of cases (%) in which medical chart documentation indicated that behavioral sleep problems were addressed by M.D. directly, via referral to sleep psychologist, and either M.D. directly and/or referral to sleep psychologist.

### 3.2. Parent-reported sleep problems (Aim 1)

All CSHQ subscale scores and the CSHQ total score for the study sample and the community control reference sample [30] are summarized in Table 2. Sample means for all subscale scores were higher than the published control reference values [30]. All subscale mean score differences were statistically significant, indicating significant sleep problems across all the subscales measuring primary medical and behavioral sleep disorders in children. The mean value of the CSHQ total score for the sample was greater than the cut-off score of 41 which indicates clinically significant sleep problems for the sample as a whole [30].

### 3.3. Prevalence of clinically significant BSD (Aim 2)

CSHQ–BSD subscale scores were examined for children with and without polysomnographically confirmed OSA (Table 3). Children in the OSA group ($n$ = 87) and the non-OSA group ($n$ = 60) did not differ with respect to all identified covariates (age ($t$[145] = 1.445; $P$ = NS); family history of excessive daytime sleepiness ($X^2$ = .230; $P$ = NS) and insomnia ($X^2$ = .456; $P$ = NS); medical history of gastroesophageal reflux ($X^2$ = .495; $P$ = NS) and asthma ($X^2$ = .001; $P$ = NS). One-way ANOVA did not reveal any statistically significant differences between mean scores on any of the CSHQ–BSD subscales (Table 3.) This finding indicates that the severity of BSD was similar for children with and without OSA. Chi-square analyses indicated that the distribution of prevalence rates for clinically significant BSD was similar (i.e., not statistically different) for the OSA and non-OSA groups, thus prevalence rates were reported for the sample as a whole (Table 4). The prevalence of specific BSD ranged from 19.1% (sleep onset delay) to 51.7% (sleep duration) for the entire sample (Table 4).

### 3.4. Comorbidity of clinically significant BSD and OSA (Aim 3)

The classification of sample subgroups based on sleep diagnoses is presented in Fig. 1. Children with comorbid OSA and BSD represented the largest diagnostic group (39.5%), followed by patients with a BSD alone (28.6%) and patients with OSA alone (19.7%).

### 3.5. Treatment of clinically significant BSD (Aim 4)

Frequency data for the management of behavioral sleep problems in the sample are summarized in Table 5. Bedtime resistance was most often addressed directly by the treating physician (53.5%), followed by sleep duration (40.8%), sleep anxiety (40.4%), and sleep onset delay (35.7%). Referrals to the sleep center psychologist were documented in 27.7% (sleep anxiety) to 41.9% (bedtime resistance) of cases with clinically significant BSD. There was no evidence of treatment documented in the medical management plan for many patients. These data indicate that (irrespective of the type of BSD) in many cases clinically significant BSD may be untreated.

## 4. Discussion

The primary purpose of this study was to examine the prevalence of clinically significant BSD in a sample of children referred for evaluation of OSA. BSD were chosen as opposed to other nonrespiratory sleep disorders because BSD are the most prevalent of the nonrespiratory sleep disorders. We chose to focus specifically on patients who were referred for OSA evaluation (in the absence of other known sleep complaints) because it was suspected that the

risk of overlooking BSD in this group was higher than children presenting with general sleep complaints. In addition, we focused on age range that presumably did not sample pre and post-"puberty" children. An examination of the extent to which sleep physicians addressed BSD (i.e., provided treatment recommendations or made a referral for behavioral treatment) within the context of an OSA evaluation was deemed clinically relevant due to anecdotal clinical observations that BSD are not routinely detected and treated in children evaluated for OSA. Overall, the study results were consistent with a priori expectations that BSD would be a clinically significant problem in children clinically referred for evaluation of OSA and that clinically significant BSD would not routinely be detected and treated during clinical care. In addition, the study findings were generally consistent with the few other studies that have examined the comorbidity of BSD and OSA in children.

### 4.1. Disrupted sleep in children clinically referred for evaluation of OSA

Although the aims of this study were focused on BSD, descriptive statistics on the full CSHQ were reported. As would be expected in a clinically referred sample of children with sleep disturbance, the CSHQ total mean score was indicative of sleep problems in the sample. It is noteworthy that the sample had statistically significant elevations on all CSHQ subscales. This finding indicates that in addition to BSD, children clinically referred for OSA evaluation are likely to present with other sleep disorder symptoms that go beyond the hallmark clinical symptoms of OSA (loud snoring; snorting and gasping for air; breathing pauses during sleep). Children in this sample were observed by their caregivers to wake frequently from sleep, experience parasomnia symptoms, and display signs of daytime sleepiness. These data are consistent with a recent study of clinically referred children with symptoms of sleep disordered breathing that were found to have a high rate of comorbid sleep problems (49%) including movement disorders, parasomnias, hypersomnia, insomnia, and circadian rhythm disturbance [27]. These results underscore the importance of using a broad based clinical evaluation that screens for both respiratory and nonrespiratory sleep disorders.

### 4.2. Prevalence of clinically significant BSD and comorbidity with OSA

It is not surprising that clinically significant BSD were identified in the sample. But the high prevalence of clinically significant BSD irrespective of the presence or absence of OSA was not fully anticipated. Our findings were consistent with prior research [21] indicating that that comorbid BSD and OSA are common. In our sample children with comorbid BSD and OSA outnumbered children with BSD alone and OSA alone. The clinical salience of this particular finding is clear in light of previous research studies demonstrating that children with comorbid OSA and BSD have greater daytime sequelae than children with either BSD or OSA alone [21,26]. Anecdotal experience within our clinical practice suggests that patients having undergone OSA evaluation are more likely to be referred for behavioral sleep medicine (psychology) services after OSA has been ruled out and sleep complaints persist that presumed to be behaviorally based. These data suggest that it is important to address BSD proactively in patients even when children have a confirmed diagnosis of OSA.

### 4.3. Treatment of clinically significant BSD

It is important to further consider the finding that an appreciable number of children with clinically significant BSD did not receive behavioral intervention for their BSD. In our center, access to behavioral sleep medicine services is not usually an issue as there is a full-time psychologist certified in behavioral sleep medicine on the sleep disorders center staff. Thus it is less likely that BSD were not addressed in the medical treatment plan due to lack of access to behavioral sleep medicine services. It more likely that the BSD were not addressed because they were deemphasized or undetected in lieu of efforts to address concerns regarding OSA. There is no means for quantifying the extent to which the CSHQ was considered during the assessment process. It is possible that the CSHQ could have been more clinically useful if the measure was formally scored. Utilization of clinical cut-off scores to identify children with clinically significant behavioral sleep problems may have improved identification and treatment of BSD. It is not certain if the observed trend for treatment of BSD is consistent with other clinical settings based solely on our findings. Future research targeting this question is warranted.

### 4.4. Significance and limitations

This study advances previous research in this area in several important ways. First, most prior research reporting comorbid OSA and BSD have either been based on anecdotal reports [23,36,37] or were conducted in school [25] or community-based general medical practices [26]. Other than one previous study conducted by Owens et al. [21], we are not aware of any other published research that has investigated the comorbidity of OSA and BSD in children who were clinically referred to a sleep disorders center for evaluation. Second, our sampling strategy that focused exclusively on children referred for sleep apnea evaluation in the absence of known or suspected behavioral comorbidities provides more specificity with respect to our findings. Third, we have gone beyond merely reporting mean score differences on measures of BSD. We took additional steps to better clarify the clinical significance of BSD by applying cut-off criteria on a measure of BSD relative to a reference control sample. Finally, we examined the extent to which physicians addressed BSD in a sample of children referred for OSA evaluation. We are not aware of any previous study that has examined physician behavior in this regard.

There are a number of study limitations that should be considered when interpreting the study findings. First, our study was designed to select a sample of children that historically has been considered to be at low risk for behavioral sleep disturbances. Our sampling strategy was developed to capture children who were suspected to have sleep disordered breathing but were not suspected to have behavioral sleep problems. Our sampling strategy relied on referral information that may have resulted in sampling bias due to inaccurate or omitted information in the referral. The potential bias due to sampling error was addressed by the clinical assessment protocol that objectively measured symptoms of sleep disordered breathing and used a well validated paper–pencil measure for assessing behavioral sleep disturbances. Second, it is highly recognized that rater bias can influence self-report measures. The CSHQ is a parent report questionnaire that is not immune to this methodological issue. Third, as previously discussed, we believe that use of a clinical cut-off score on the CSHQ to identify clinically significant BSD is strength of this study; however, it should be noted that this criterion does not imply a formal diagnosis of behavioral insomnia based on diagnostic criteria. In addition, although the four CSHQ–BSD subscales measure different types of sleep behaviour, the subscales do not necessarily represent four distinct disorders/diagnoses. Fourth, the methodology we used for coding physician behavior with respect to treating clinically significant BSD relied on medical chart documentation. There may have been cases where BSD had been addressed by the treating physician but was not identified during the medical chart coding procedures due to lack of strict documentation guidelines for coding behavioral treatment. Finally, care should be taken when interpreting this study with respect to

generalizability because we sampled a referral population with a specific clinical concern.

## 5. Conclusions

Overall, these findings confirm the necessity of conducting comprehensive sleep evaluations that screen for respiratory and nonrespiratory disorders. In fact, the data suggest that an emphasis on detecting clinically significant BSD is warranted. This may be accomplished by paying close attention to concerns related to the bedtime transition, parent–child interactions at bedtime, prolonged sleep onset, sleep related anxiety, and inadequate sleep. Utilization of clinical cut-off criteria on the CSHQ may prove helpful for identifying significant BSD in the clinical setting. The study data confirm the reality that ruling out OSA is rarely the end point of a sleep evaluation.

Closer attention to the presence and impact of comorbid BSD should improve diagnostic accuracy in children that have historically been considered to be at low risk for clinically significant BSD. In addition, because there is research support for the effective treatment of BSD [12,15,38–41], consistent identification of BSD followed by empirically supported behavioral treatment should yield improved treatment outcomes. It is evident that sleep medicine physicians must be proficient in formulating and overseeing both medical and behavioral treatments or have access to a referral source that provides behavioral sleep medicine services. It is our experience that children with complex sleep disorders are best cared for within the context of a multidisciplinary team. As the study demonstrates, however, even within the context of a multidisciplinary sleep disorders clinic, clinically significant BSD may go undetected and untreated. Thus, sleep medicine clinicians must continually strive to improve clinical care by integrating new research evidence into assessment and treatment practices.

*Disclosure statement*

The authors on this project have no disclosures regarding conflicts of interest. The project was completed in the absence of any related (1) research grant of contract support administered through an academic or research institute; (2) personal compensation through contract, grants, honoraria, fees, or salary, and (3) personal financial investment, including ownership and equity or other financial holdings.

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