Inability of Clinical History to Distinguish Primary Snoring From Obstructive Sleep Apnea Syndrome in Children*

John L. Carroll, MD; Susanna A. McColley, MD; Carole L. Marcus, MBBCh; Shelly Curtis, RN; and Gerald M. Loughlin, MD

Study objective: To determine whether primary snoring (PS) could be distinguished from childhood obstructive sleep apnea syndrome (OSAS) by clinical history.

Design: Retrospective study of clinical history of 83 children with snoring and/or sleep disordered breathing who were referred for polysomnography.

Setting: Tertiary referral center; pediatric pulmonary sleep apnea clinic.

Measurements: We evaluated the ability of a clinical obstructive sleep apnea (OSA) score and other questions about sleep, breathing, and daytime symptoms to distinguish PS from OSAS in children. Parents were asked about the child's snoring, difficulty breathing, observed apnea, cyanosis, struggling to breathe, shaking the child to "make him or her breathe," watching the child sleep, snoring, and loudness of snoring, and daytime symptoms such as excessive daytime sleepiness (EDS).

Results: Based on polysomnography results, 48 patients were classified as PS and 35 as OSAS. Peak end-tidal CO₂ (49±3.2 vs 55±8.2 [SD] mm Hg); lowest arterial oxygen saturation measured by pulse oximetry (95±1.9 vs 82±14%); and apnea/hypopnea index (0.27±0.3 vs 8.4±6 events/h) indicated that the diagnostic criteria for PS versus OSA were reasonable. There were no differences between PS and OSA patients with respect to age, sex, race, failure to thrive, obesity, history of EDS, snoring history, history of cyanosis during sleep, or daytime symptoms except for mouth breathing. There were no significant differences in sleep variables between PS patients and those with any severity of OSAS. The OSA score misclassified about one of four patients. Comparing PS and OSA patients, significant findings were daytime mouth breathing (61 vs 85%; p=0.024); observed apnea (46 vs 74%; p=0.013); shaking the child (31 vs. 60%; p=0.01); struggling to breathe (55 vs 89%; p=0.003); and afraid of apnea (71 vs 91%; p=0.028). However, none of these were sufficiently discriminatory to predict OSAS.

Conclusion: We conclude that PS in children cannot be reliably distinguished from OSAS by clinical history alone.

(AI=apnea/hypopnea index; EDS=excessive daytime sleepiness; OSA=obstructive sleep apnea; OSAS=obstructive sleep apnea syndrome; PS=primary snoring; PSG=polysomnography; UARS=upper airway resistance syndrome)

Key words: child; clinical history; obstructive sleep apnea syndrome; primary snoring; questionnaire; sleep; sleep-disordered breathing; snoring

Although precise incidence figures are not available, current evidence suggests that childhood obstructive sleep apnea syndrome (OSAS) affects about 1 to 2% of children.1,2 It is characterized by snoring or noisy breathing during sleep associated with some combination of hypoxemia, hypercapnia, sleep disturbances, or daytime symptoms such as mouth breathing, abnormal behavior, and excessive daytime sleepiness (EDS).3 Children with OSAS may be male or female, and they are usually referred for evaluation because of snoring, noisy breathing, or difficulty breathing during sleep. This contrasts markedly with adult-type OSAS in which patients are often obese males, and the main presenting complaint is EDS.3

The clinical diagnosis of childhood OSAS is complicated by the fact that the incidence of habitual snoring in children, that is, snoring on most nights, is reported to be 7 to 9%,1,2,4 Thus, most children with habitual snoring have primary snoring (PS), characterized by snoring without associated hypoxemia, hypercapnia, sleep disruption, or daytime symptoms.3 Only a subgroup of habitually snoring children have OSAS. Although PS is a benign condition that usually does not require treatment, childhood OSAS, if left untreated, can lead to failure-to-thrive, cor pulmonale,
developmental delay, and even death. For the child with habitual snoring, it is essential that the correct diagnosis be made so that those with childhood OSAS can receive appropriate treatment (usually adenotonsillectomy) and so that those with primary snoring can avoid unnecessary surgery.

Because of the time and expense of using full polysomnography (PSG) to diagnose childhood OSAS (up to $1,500 per study), Brouillette et al. investigated whether OSAS in children could be diagnosed without it. Using a clinical scoring system (obstructive sleep apnea [OSA] score) based on three questions about breathing during sleep, they concluded that children with no OSAS or severe OSAS could be identified by history alone. In addition, it was proposed that PSG was only necessary for children with mild-moderate OSAS, in which the OSA score failed to classify them as clearly normal or clearly having severe OSAS.

In the study by Brouillette et al., non-OSAS control subjects consisted of normal children seen in a general pediatric clinic. However, in a pediatric sleep apnea clinic setting, most children undergoing evaluation for possible OSAS are referred because of snoring or difficulty breathing during sleep. Whether the OSA score would distinguish PS from OSAS in this setting, in which all children snore, is unclear. The aims of this study were, therefore: (1) to determine if the OSA score is applicable in a pediatric sleep apnea clinic setting, (2) to determine if a reformulation of the OSA score, expanded to include other features of PS and OSAS, would be discriminative in this setting, (3) to determine the reported incidence of symptoms in children with PS versus OSAS, and (4) to determine if nonrespiratory polysomnographic findings differ in primary snorers versus children with OSAS.

**METHODS**

**Patient Selection**

At the beginning of their visit to our Pediatric Sleep and Breathing Disorders Clinic, as part of the clinical evaluation, a questionnaire inquiring about the child’s sleep and breathing during sleep was filled out by the parents of all children referred for evaluation of possible OSAS. Each child was then studied using full overnight PSG in order to detect sleep-related upper airway obstruction, as discussed later on. The PSGs were scored without knowledge of questionnaire results. Children with significant medical problems, previous upper airway surgery, or craniofacial abnormalities were excluded. Eighty-three children referred for evaluation of possible OSAS over a 14-month period met the criteria for inclusion. Based on PSG results, children were separated into two groups as follows: group 1, children with PS (snorers without OSAS on PSG), and group 2, children with OSAS (patients with OSAS diagnosed by PSG).

**Obstructive Sleep Apnea Score**

The OSA score was based on the answers to three questions as follows: (a) How often would you say your child has difficulty breathing when he or she is sleeping? (0=never, 1=occasionally; 2=frequently, 3=constantly). (b) Does your child stop breathing when he or she is asleep? (0=no, 1=yes). (c) How often would you say your child snores? (0=never, 1=occasionally, 2=frequently, 3=constantly). The answers to questions a through c were then used to calculate the OSA score according to the equation described by Brouillette et al. as follows: OSA score = $1.42(a) + 1.41(b) + 0.71(c) - 3.73$.

<table>
<thead>
<tr>
<th>Questions:</th>
<th>Choices</th>
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<tbody>
<tr>
<td>Do you ever see your child stop breathing during sleep?</td>
<td>No</td>
</tr>
<tr>
<td>Do you ever see your child struggling to breathe during sleep?</td>
<td>No</td>
</tr>
<tr>
<td>When your child is asleep, do you ever shake him or her to make him or her start breathing again?</td>
<td>No</td>
</tr>
<tr>
<td>Do you ever see your child’s lips or skin turn blue or purple during sleep?</td>
<td>No</td>
</tr>
<tr>
<td>Do you watch your child while he or she is asleep at night, afraid about his or her breathing?</td>
<td>Never, Rarely</td>
</tr>
<tr>
<td>How loud is the snoring?</td>
<td>Mild, quiet</td>
</tr>
<tr>
<td>How often does your child have a sore throat?</td>
<td>Never, Rarely</td>
</tr>
<tr>
<td>Does he or she complain of morning headaches?</td>
<td>Never</td>
</tr>
<tr>
<td>Is your child a daytime mouth breather?</td>
<td>Never</td>
</tr>
<tr>
<td>How big of a problem does your child have with sleepiness during the daytime?</td>
<td>None, Slight</td>
</tr>
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</table>
Expanded Obstructive Sleep Apnea Questionnaire

Experience evaluating children referred for possible OSAS prior to this study suggested that other aspects of the clinical history not covered by the OSA score may be important. We therefore expanded the questionnaire to include more questions about difficulty breathing during sleep and daytime symptoms (Table 1). All questionnaires were administered by the same interviewer (S.C.).

Polysomnography

Polysomnography was performed overnight in a quiet, comfortable, darkened room, with one parent sleeping in the same room with the child. No sleep deprivation or sedation was used to induce sleep. During PSG, the following signals were recorded continuously on a physiologic recorder (Grass Model 78-D; Grass Instruments; Quincy, Mass):

1. Sleep staging using two leads (C3-A2/C4-A1) for EEG, right and left electro-oculograms, and submental electromyogram.
2. Chest and abdominal motion was measured using mercury-filled strain gauges.
3. An ECG was monitored.
4. Arterial oxygen saturation was monitored continuously using a Nellcor N-100 or N-1000 oximeter operating in mode 2 (fast [Nellcor; Hayward, Calif]).
5. The pulse oximeter waveform was recorded in order to detect motion artifact.
6. Airflow was semiquantitatively monitored using a three-bead thermistor positioned over nostrils and the mouth (Somnitec; Van Nuys, Calif).
7. In some patients end-tidal CO2 was monitored continuously via a nasal cannula using a Nellcor N-1000 (Nellcor; Hayward, Calif).
8. The entire polysomnogram of each child was audiotaped and videotaped using infrared lights. Each child was continuously monitored by a technician trained in pediatric PSG.

Polysomnography Scoring

Sleep was staged according to standard guidelines. For several reasons, a significant obstructive event duration was defined as at least 2 respiratory cycle times rather than the adult standard of 10 s. The higher oxygen consumption-oxygen supply ratio of young children compared with adults, combined with a decrease in functional residual capacity during sleep, may lead to rapidly developing hypoxemia during brief apneas. Also, due to the faster respiratory rates of children, 10 s may represent 2 to 3 missed breaths in an adult but 5 to 6 missed breaths in a young child. Obstructive apnea was defined as absent oronasal airflow in the presence of continued respiratory effort for at least 2 respiratory cycle times, accompanied by a 4% or greater decrease in arterial oxygen saturation. Obstructive hypopnea was defined as a decrease of 50% or more in the amplitude of the oronasal thermistor signal, with maintained respiratory effort, for at least 2 respiratory cycle times and accompanied by a 4% or greater decrease in arterial oxygen saturation. An apnea/hypopnea index (AHI) was calculated as follows: (No. of obstructive apneic events+No. of obstructive hypopnea events)/hours of sleep time=No. of obstructive respiratory events per hour of sleep time.

Data Analysis

Primary Snoring vs Obstructive Sleep Apnea. For initial data analysis, based on the AHI, patients were classified into two groups as follows: group 1, PS-AHI <1/h; group 2, childhood OSA-AHI ≥1/h or more. In order to determine if different PSG diagnostic criteria improved discrimination, data were re-analyzed in several ways as follows: (1) PS group, defined as aforementioned, was also compared with OSAS patients with an AHI ≥5/h, or AHI ≥10/h; and (2) PS group, defined as no hypoxemia, no hypercarbia, and AHI=0, was compared with OSAS patients with AHI ≥1/h, AHI ≥5/h; or AHI ≥10/h. Polysomnography data were also analyzed for correlations with age.

Obstructive Sleep Apnea Score: After classifying children as having PS versus childhood OSAS using the criteria of PS-AHI <1/h and childhood OSA-AHI ≥1/h or more, we then examined whether the OSA score would have correctly classified each child as not having OSAS or having severe OSAS. According the guidelines of Brouillette et al, patients were classified based on their OSA score into three groups: (1) OSA score of < -1=no OSA; (2) OSA score of -1 to 3.5=indeterminate; (3) OSA score of >3.5=OSAS. Based on PSG results, the proportions of subjects correctly or incorrectly classified by the OSA score were determined.

Expanded Clinical History: After classifying subjects as having PS vs childhood OSAS based on PSG results as described previously, nominal data from the expanded questionnaire (Table 1) were analyzed using the Fisher’s exact test for 2×2 tables, the Pearson χ2 test for larger tables, and odds ratios obtained from multiple logistic regression (Crunch 4.0, August 1993 version; Crunch Software; Oakland, Calif). The best multivariate model was first tested on the dataset from which it was derived to determine how well it would have predicted the diagnosis of PS or OSAS. The data were then randomly stratified into two subsets with equal proportions of PS and OSAS patients. Using logistic regression analysis to derive the best model from one subset, it was then applied to the other data subset by calculating probabilities of having PS vs OSAS for each patient. The optimal classification criterion was determined (cutpoint that minimizes false classification), and patients in the second data subset were classified as having PS vs OSAS. Agreement between actual group membership and predicted group membership was tested using the kappa statistic (Crunch 4.0; Crunch Software; Oakland, Calif). The model was not tested further in this study, since no combination of reported symptoms was found to yield acceptable predictions. A prediction error of 10% or less was considered acceptable. Means are presented as mean±1 SD. A probability value of less than 0.05 was considered significant.

Results

Eighty-three children met the criteria for inclusion in the study, ranging in age from 5.4 months to 14.8 years. Mean age was 5.6±3.4 years in group 1 and 4.3±2.4 in group 2 (NS). Fifty-eight percent of subjects were male, 26 (31%) were white, 56 (67.5%) were African-American, and 1 child was Asian. Obesity (weight greater than, 95th percentile for age) was present in 21 (26%) subjects, and 3 (3.7%) were underweight (less than 5th percentile) for age. All patients were otherwise well, except for snoring or OSAS at the time of the study.

Polysomnographic data are shown in Table 2. It will be noted that the AHI of the PS group was not zero. This was because 22 of 48 patients diagnosed with PS (group 1) had at least 1 episode of upper airway obstruction during sleep. However, these obstructive episodes occurred less than once an hour and only 1 patient (with an isolated desaturation to 89%) had an oxyhemoglobin desaturation level of less than 92%. This is consistent with the polysomnographic data of Marcus et al for normal children, which showed that 49 of 50 normal children had nadir oxygen saturation values above 90% during sleep but also reported an isolated desaturation in one child to 89%. In order to
determine whether PS patients with some obstructive events (0 < AHI < 1) were affecting the results, we reanalyzed the sleep variables listed in Table 2 after restricting the PS group to children with no obstructive events (AHI = 0 [n = 26]); the results were the same as for the entire PS group (n = 48). Measures of sleep efficiency and sleep architecture did not differ significantly between primary snorers and subjects with obstructed breathing of any severity (Table 2). Although it was not the focus of this study, we found, as expected, that the number of rapid eye movement periods per night (p = 0.0001), the percentage of total sleep time spent in rapid eye movement sleep (p = 0.013), and the number of movements per hour (p = 0.044) declined significantly with age. Other variables shown in Table 2 did not correlate significantly with age.

Twenty-seven out of 48 patients who were diagnosed by PSG as having PS had indeterminate OSA scores (requiring PSG for diagnosis [Fig 1, group 1]). Of the remaining 21 subjects, the OSA score classified 15 as not having OSA (OSA score of < -1) and 6 subjects as positive for OSA (OSA score of > 3.5). Therefore, the OSA score would have classified 6 of 21 (29%) as having OSAS when polysomnography showed only PS (Fig 1, group 1).

Twenty-one of 35 patients diagnosed by PSG with childhood OSAS had indeterminate OSA scores and, according to the guidelines for using the OSA score,1 would have required PSG for diagnosis (Fig 1, group 2). Of the remaining 14 subjects with OSA scores of < -1 or > 3.5, 11 (79%) were correctly classified as having OSAS and 3 (21%) were erroneously classified as not having OSAS (Fig 1, group 2). Therefore, according to the guidelines for using the OSA score,1 35 of 83 patients would have been diagnosed without PSG. Based on results of full PSG, of those 35 patients with OSA scores of < -1 or > 3.5, 9 or 26% would have been misdiagnosed.

Historical features obtained using the expanded childhood OSAS questionnaire are shown in Table 3. There were no significant differences between the PS and OSAS subjects with respect to failure to thrive, obesity, snoring characteristics, or daytime symptoms except for mouth breathing (Table 3). Parental observation of cyanosis during sleep was uncommon in both groups. Parental observations of obstructive apnea and struggling to breathe during sleep were significantly greater in the OSAS group but still present in 46 to 58% of the PS group (Table 3). Intervention by a parent to “make” the child breathe was twice as frequent in the OSAS group but was still reported by about one third of the parents of children in the PS group. Although 91% of parents of children with OSAS watched the child sleeping, afraid about his or her breathing, so did about two thirds (71%) of the parents of children with PS.

Multiple logistic regression yielded the best model with 3 variables, observed apnea, observed struggling,
and watching the child sleep (p=0.0016). However, when this model was tested for its ability to predict which children had PS vs OSAS, it performed no better than the OSA score. Even when tested on the dataset from which it was derived, the best overall correct prediction rate was only 67%. When derived from a random stratified subset of the data (50%) and applied to a second random data subset (50%), the logistic model performed better than chance alone (kappa=-0.118; p=0.01). However, only 68% of patients predicted to have PS and 78% of patients predicted to have OSAS were correctly identified.

The data were re-analyzed using an AHI of >5 or 10 to classify patients as having OSA. The outcome, with respect to sleep (Table 2) or reported symptoms was similar. As expected, when the AHI cutoff was raised, the proportion of reported symptoms (eg, observed apnea, struggling, shaking) increased in the OSA group. However, the predictive value of these symptoms was not enhanced, due to the high reported frequency of these symptoms in the PS group.

**Discussion**

This study examined, in a clinical setting in which children were referred for snoring or symptoms of sleep-disordered breathing or both, whether primary snoring could be distinguished, by history alone, from obstructive sleep apnea. The results indicate that in patients with PS vs OSAS (1) most of the patients, even when referred because of snoring or sleep-disordered breathing, did not meet even minimal PSG criteria for

| Table 3—Historical Features of Primary Snoring (AHI <1) vs Childhood (AHI ≥1)* |
|----------------------------------|--------------|----------------|-----------------|-----------------|
|                                  | PS, %        | OSAS, %        | χ^2 or Fisher’s Exact Test | Odds Ratio      | 95% CI          |
| General                          |              |                |                             |                 |                |
| Failure to thrive (82)           | 4            | 3              | NS                           | 0.66            | 0.06 7.60      |
| Obesity (81)                     | 28           | 23             | NS                           | 0.75            | 0.27 2.08      |
| Daytime symptoms                 |              |                |                               |                 |                |
| No history of EDS (68)           | 71           | 74             | NS                           | 1.13            | 0.40 3.53      |
| Slight EDS                       | 12           | 19             | NS                           | 1.64            | 0.42 6.30      |
| Moderate/severe EDS              | 17           | 7              | NS                           | 0.39            | 0.07 2.03      |
| Morning headache more than once a week (46) | 23           | 0              | NS                           | ...             | ...            |
| Sore throat >6 times per year (52) | 22           | 5              | NS                           | 0.19            | 0.02 1.66      |
| Runny nose frequently or constantly (83) | 33           | 31             | NS                           | 0.91            | 0.36 2.33      |
| Frequent or constant mouth breathing (80) | 61           | 85             | p=0.024                       | 3.73            | 1.22 11.41     |
| Snoring                          |              |                |                               |                 |                |
| Snoring loud or extremely loud (83) | 83           | 91             | NS                           | 1.75            | 0.47 6.50      |
| Snores “most nights” (83)        | 96           | 97             | NS                           | 4.00            | 0.43 37.42     |
| Parent’s observations            |              |                |                               |                 |                |
| Observed cyanosis during sleep (83) | 4            | 6              | NS                           | 3.41            | 0.34 34.17     |
| Observed obstructive apnea during sleep (83) | 46           | 74             | p=0.013                       | 3.34            | 1.32 8.80      |
| Observed child struggling to breathe during sleep (83) | 58           | 89             | p=0.003                       | 5.54            | 1.67 18.18     |
| Shakes child to make him or her breathe during sleep (83) | 31           | 60             | p=0.01                        | 3.30            | 1.33 8.21      |
| Watches child sleeping, afraid about breathing (83) | 71           | 91             | p=0.028                       | 4.39            | 1.15 16.73     |

*Numbers of subjects are in parentheses; NS=not significant.
a diagnosis of OSAS; (2) the OSA score was not able to discriminate accurately between children who have only snoring versus children with snoring and sleep-related airway obstruction; (3) most reported daytime symptoms or other clinical features such as obesity or failure to thrive were not significantly different; (4) reported snoring frequency or intensity were not significantly different; (5) difficulty breathing during sleep was more frequently reported by parents of OSAS patients but differences were not predictive; and (6) sleep efficiency and architecture did not differ significantly. These findings suggest that evaluation of the snoring child for possible OSAS must include some objective method of detecting upper airway obstruction during sleep; clinical history alone is not adequate to distinguish childhood PS from OSAS.

Critique of Methods

Although there are no published reports on the night-to-night variability of symptoms in childhood OSAS, experience suggests that they may vary from night to night. Therefore, it is possible that some of our subjects were misclassified by PSG, since it only measures sleep and breathing during one night. Some children with an AHI of <1, classified as having PS, may have been OSA patients who are worse at home or on other nights. However, we believe this is unlikely, since 22 of the PS group had no obstructive events at all and showed essentially the same frequency of reported symptoms as the overall PS group. In the PS patients with an AHI of 0, reported symptom frequencies were as follows (n=22): cyanosis, 5%; observed apnea, 40%; observed struggling to breathe, 50%; shaking child to stimulate breathing, 30%; watching child because afraid about breathing, 55%; snoring loud or extreme, 75%; and snoring most nights, 95%. These frequencies, in children with no obstructive events, are very similar to frequencies of reported symptoms in the PS group overall (Table 3). Re-analysis of the data using an AHI of 0 to distinguish PS from OSAS did not change the outcome. Thus, the frequency of symptoms in the PS group appears to represent the clinical picture of PS and does not appear to be skewed by misclassified OSA children.

It is possible that some of the childhood PS group may have been children with obstructive hypoventilation and little or no OSA per se. Obstructive hypoventilation, defined as partial upper airway obstruction leading to hypercapnia, may occur in the absence of complete obstructive apnea. Three of the 48 PS patients had isolated peak end-tidal CO$_2$ values above 52 mm Hg (54, 54, and 56 mm Hg) and at the time of this study end-tidal CO$_2$ was not always measured for the entire sleep period in our laboratory. However, none of these patients had oxygen desaturations below 94%. In addition, when the analysis is restricted to patients with a peak end-tidal CO$_2$ of less than 52 mm Hg, sleep variables and frequencies of reported symptoms did not differ from those of the PS group overall. Therefore, there is no evidence that our PS group was significantly affected by misclassification of patients with missed obstructive hypoventilation.

Marcus et al$^7$ have shown that normal children may have up to one obstructive apnea per hour. Therefore, based on normal values for children, we classified patients without hypoxemia and less than 1 obstructive event per hour as having PS. Analysis of the data using an AHI of zero instead of AHI <1 did not change the outcome of the analysis for sleep variables or historical features. On the other hand, it is also possible that our OSAS criterion of AHI ≥1 event per hour was too low. However, reanalysis using an AHI criterion of ≥5 or ≥10 events per hour also did not affect the outcome of the analysis for sleep variables or historical features. Indeed, the greatest number of significant differences between PS and OSA groups was obtained using an AHI criterion of ≥1 event per hour.

Diagnosis of Childhood Obstructive Sleep Apnea Syndrome by Clinical History

In the 1984 study by Brouillette et al,$^3$ the control group consisted of normal children from general pediatric clinics. The OSAS patients, on the other hand, consisted almost entirely of snoring children and all were diagnosed by polysomnography.$^3$ Snoring was reported in 96% (22 of 23) of the patient group, but only 9% (4 of 45) of the control group in that study.$^3$ Therefore, although the study of Brouillette et al$^3$ demonstrated that some children with severe OSAS can be distinguished from some normal control subjects by history alone, the question remained whether the OSA score would work in a clinical setting in which nearly all children are snorers. In our experience, obstructed breathing during sleep in children is usually suspected because of loud snoring, and most children referred for clinical evaluation of suspected OSAS have a history of snoring. The results of the present study demonstrate that the OSA score did not perform adequately in this clinical setting, most likely because it was not designed to discriminate between snorers without OSA and snoring children with OSAS. Table 4 compares our primary snorers (group 1) with the normal control group in the study of Brouillette et al$^3$ (Table 4). Our PS group was considerably more symptomatic compared with the control children in their study,$^3$ which most likely accounts for the failure of the OSA score to work adequately in our clinical setting (Table 4).

Although the OSA score, as described by Brouillette et al,$^3$ correctly predicted the polysomnographic diagnosis in about three fourths of cases falling outside of the indeterminate range (Fig 1), we believe that mis-
diagnosing one fourth of cases is still unacceptable. In our hands, the OSA score had a sensitivity and specificity of 73 and 83%, respectively. Closer inspection of the nine cases misclassified by the OSA score did not reveal an explanation for their incorrect assignment. In every one of these cases, the polysomnographic results were unequivocal. Four of the six patients misclassified as “positive” (OSA score of >3.5) by history had AHIs of 0 and the other two had AHIs of 0.2 and 0.5 events per hour. The 3 patients misclassified as “negative” (OSA score of <1) by history had AHIs of 3.3, 4.6, and 36 events per hour, significant oxyhemoglobin desaturation, and 2 of 3 had prolonged hypercarbia. Sleep architecture, recording times, and sleep efficiency were not significantly different in patients with false-positive versus false-negative OSA scores.

Because it would be desirable to have an effective screening tool based on clinical history, we attempted to improve the clinical OSA score by expanding it to include other symptoms (Table 3). However, univariate analysis showed that general features, reported daytime symptoms, snoring frequency, snoring intensity, and cyanosis during sleep were not significantly different between snorers with versus without OSA (Table 3). The historical features that were statistically significantly different between the PS and OSA groups were similar to those included in the OSA score except for snoring1 (Table 3). Reported snoring drops out of our analysis because its prevalence was so high in both groups. Logistic regression analysis revealed that no combination of reported symptoms was sufficiently predictive for use in the clinical setting in which nearly all children are snorers. Our best model, consisting of the variables observed apnea, snoring, and watching the child sleep (afraid about breathing), when applied to the dataset from which it was derived, would have correctly identified 92% of the OSA patients but misclassified approximately 50% of primary snorers as having OSA. We did not attempt to derive a discriminant function because of the extensive overlap of reported symptoms in the two groups.

Clinical Features and Symptoms of Primary Snoring vs Obstructive Sleep Apnea Syndrome in Children

Although reported symptoms were not of discriminative value, several symptoms were significantly more common in the OSA group (Table 3). While ~90% of parents of OSA children perceive that their child is struggling to breathe and watch the child during sleep afraid about his or her breathing, it is noteworthy that more than 50% of parents of children with PS have the same perceptions. Forty-six percent of parents of primary snorers reported seeing obstructive apnea and 31% even reported shaking the child during sleep to stimulate breathing (Table 3). Even when restricted to the PS group in which AHI is 0 on PSG, 40% of parents reported observed apnea. Assuming that PSG correctly classified children with PS, these data suggest that parents are frightened by both PS and OSA and that it is very difficult for parents to tell the difference.

The reasons why many parents of primary snorers report dramatic symptoms, such as observing the child struggling to breathe, are probably related to the high degree of symptom overlap between PS and OSAS. If PS and OSAS lie on a continuum, then the distinction between one versus the other is a matter of degree, not a matter of qualitative differences in symptomatology. Thus, it is likely that children with PS are accurately perceived by parents as having intermittent obstructive apnea, “struggling” to breathe during sleep, and snoring loudly. In addition, our experience is that parental perceptions of snoring and parental thresholds for concern are quite variable, possibly accounting for some of the dramatic reports of symptoms from parents of children with only mild PS on PSG.

One striking finding was that standard measures of sleep architecture were little affected by the presence of OSA in these children. Table 2 shows that children with PS or OSA of any severity showed no differences in measures of sleep quality. We did not measure daytime sleepiness nor did we measure microarousals, leaving open the possibility that more subtle measures of sleep quality would show differences. However, data from our laboratory10 and from other studies of children with OSA11 have shown that children may have repeated episodes of OSAS without showing EEG arousal. In addition, the lack of EDS in nearly 80% of the OSA group suggests that their sleep quality is adequate. Thus, sleep appears to be less disrupted by OSA in children than it is in adults, possibly accounting for the lack of differences in sleep variables and daytime symptoms.

Daytime symptoms and general features such as failure to thrive (FTT) or obesity did not differ significantly between PS and OSA patients (Table 3). A history of EDS, which is a hallmark of adult OSAS, was not reported by 71 and 74% of parents of PS and OSA children, respectively. Moderate or severe EDS was only reported by 7% of the OSA subjects (Table 3). These data suggest that a history of EDS in snoring

Table 4—Comparison of the Control Group From the Study of Brouillette et al1 vs the Primary Snoring Group (Group 1) in the Present Study

<table>
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<tr>
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<th>Control Group in Study of Brouillette et al1</th>
<th>PS Group in Present Study Group 1</th>
</tr>
</thead>
<tbody>
<tr>
<td>Snoring frequently or constantly, %</td>
<td>9</td>
<td>90</td>
</tr>
<tr>
<td>Stops breathing during sleep, %</td>
<td>5</td>
<td>46</td>
</tr>
<tr>
<td>Difficulty breathing during sleep frequently or constantly, %</td>
<td>2</td>
<td>42</td>
</tr>
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</table>
children is a feature of PS or adenotonsillar hypertrophy and is not necessarily due to the presence of OSA. We did not perform any tests of daytime sleepiness and therefore could not determine whether the history given by parents was accurate. In any case, parental perceptions of EDS do not appear to separate PS from snoring with OSAS in children.

Signs of adenotonsillar hypertrophy, except daytime mouth breathing, occurred to a similar degree in both groups (Table 3). Although significantly more parents in the OSA group reported daytime mouth breathing, it was also reported by more than half of the PS group parents. The frequency of daytime symptoms reported by our PS group is similar to that of the OSA group in the study of Brouillette et al. This strongly suggests that clinical features, such as daytime mouth breathing, upper respiratory congestion, snoring, etc., are features of adenotonsillar hypertrophy and not of OSAS per se. These features are clearly not predictive of OSAS.

The data in this study additionally support the concept of the upper airway resistance syndrome (UARS) in children described by Downey et al. Children with UARS have snoring with increased upper airway resistance during sleep and exhibit symptoms, such as restlessness, behavioral problems, poor school performance, and EDS, but do not meet the criteria for a polysomnographic diagnosis of OSAS. Patients with UARS appear to lie on a continuum between PS and OSAS and exhibit a great deal of symptom overlap with PS and OSAS. Of the 48 patients in our PS group, 5 patients (ages 6 to 12 years) had a history of moderate or severe EDS, and 3 of these also had histories of school problems. Therefore, a small number of our PS patients may have had UARS. Although the number was too small for statistical analysis, sleep variables were similar for these patients compared with those of other PS and OSAS patients. Further study is needed to determine the best means of identifying children with UARS.

Our findings with respect to historical features of childhood OSAS are similar to those reported by Leach et al for 93 children of similar ages presenting to an otolaryngology clinic for evaluation. They divided patients in a similar manner into PS versus OSAS groups, although their precise criteria for diagnosis of OSAS were not specified. Results resembled ours in that most of the children presenting to their clinic did not show OSA on PSG, and historical features were not significantly different between children without OSA and children exhibiting OSA on PSG. In agreement with our results, Leach et al also found that the incidence of EDS was low (~13 to 17%) and equal in the two groups. Unlike our patients, in which cyanosis was a rarely reported symptom, Leach et al reported cyanosis in about 15% of patients in both groups.

Implications

Past descriptions of the child with OSAS have included obesity, EDS, morning headaches, daytime mouth breathing, loud snoring on most nights, observed apnea, and difficulty breathing during sleep. However, our data indicate that this constellation of symptoms also applies to the child with PS. This is not surprising because PS and OSA are thought to lie on a continuum, from partial sleep-related upper airway obstruction without significant hypoxemia or hypercarbia (PS) to obstructive hypoventilation and apnea with hypoxemia and CO2 retention (OSAS). In addition, our data showing relatively normal sleep architecture and no history of EDS in most patients provide additional support for the view that childhood OSAS differs in important respects from adult-type OSAS.

Most general pediatricians in the United States do not directly order PSG for their patients. In our experience, children are referred to a pediatric sleep center, usually because of snoring or perceived difficulty breathing during sleep or both. This would appear to be a reasonable practice, because, as shown in Table 2 and by numerous other studies, nearly all patients with OSA snore. In other words, if a child does not exhibit some snoring during sleep, OSAS is very unlikely. However, the converse is not true; if a child does exhibit snoring, he or she may have only PS or may have OSA. Since the incidence of snoring in children is about 7 to 9%, while the incidence of OSAS in children is estimated to be about 1 to 2%, most snoring children would not have OSAS. Therefore, accurate diagnosis of PS versus OSAS is essential in order to avoid unnecessary surgery or other intervention in children with PS. Our data indicate that the clinician (including pediatric pulmonologists, otolaryngologists, and pediatric surgeons) evaluating a child with snoring cannot, at the present time, reliably distinguish PS from OSA without some type of measurement of breathing during sleep.

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REFERENCES


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