

## Technical Report: Diagnosis and Management of Childhood Obstructive Sleep Apnea Syndrome

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**ABSTRACT.** *Objective.* This technical report describes the procedures involved in developing the recommendations of the Subcommittee on Obstructive Sleep Apnea Syndrome in children. The group of primary interest for this report was otherwise healthy children older than 1 year who might have adenotonsillar hypertrophy or obesity as underlying risk factors of obstructive sleep apnea syndrome (OSAS). The goals of the committee were to enhance the primary care clinician's ability to recognize OSAS, identify the most appropriate procedure for diagnosis of OSAS, identify risks associated with pediatric OSAS, and evaluate management options for OSAS.

*Methods.* A literature search was initially conducted for the years 1966–1999 and then updated to include 2000. The search was limited to English language literature concerning children older than 2 and younger than 18 years. Titles and abstracts were reviewed for relevance, and committee members reviewed in detail any possibly appropriate articles to determine eligibility for inclusion. Additional articles were obtained by a review of literature and committee members' files. Committee members compiled evidence tables and met to review and discuss the literature that was collected.

*Results.* A total of 2115 titles were reviewed, of which 113 provided relevant original data for analysis. These articles were mainly case series and cross-sectional studies; overall, very few methodologically strong cohort studies or randomized, controlled trials concerning OSAS have been published. In addition, a minority of studies satisfactorily differentiated primary snoring from true OSAS. Reports of the prevalence of habitual snoring in children ranged from 3.2% to 12.1%, and estimates of OSAS ranged from 0.7% to 10.3%; these studies were too heterogeneous for data pooling. Children with sleep-disordered breathing are at increased risk for hyperactivity and learning problems. The combined odds ratio for neurobehavioral abnormalities in snoring children compared with controls is 2.93 (95% confidence interval: 2.23–3.83). A number of case series have documented decreased somatic growth in children with OSAS; right ventricular dysfunction and systemic hypertension also have been reported in children with OSAS. However, the risk growth and cardiovascular problems cannot be quantified from the published literature. Overnight polysomnography (PSG) is recognized as the gold standard for diagnosis of OSAS, and there are currently no satisfactory alternatives. The diagnostic accuracy of

symptom questionnaires and other purely clinical approaches is low. Pulse oximetry appears to be specific but insensitive. Other methods, including audiotaping or videotaping and nap or home overnight PSG, remain investigational. Adenotonsillectomy is curative in 75% to 100% of children with OSAS, including those who are obese. Up to 27% of children undergoing adenotonsillectomy for OSAS have postoperative respiratory complications, but estimates are varied. Risk factors for persistent OSAS after adenotonsillectomy include continued snoring and a high apnea-hypopnea index on the preoperative PSG.

*Conclusions.* OSAS is common in children and is associated with significant sequelae. Overnight PSG is currently the only reliable diagnostic modality that can differentiate OSAS from primary snoring. However, the PSG criteria for OSAS have not been definitively validated, and it is not clear that primary snoring without PSG-defined OSAS is benign. Adenotonsillectomy is the first-line treatment for OSAS but requires careful postoperative monitoring because of the high risk of respiratory complications. Adenotonsillectomy is usually curative, but children with persistent snoring (and perhaps with severely abnormal preoperative PSG results) should have PSG repeated postoperatively. *Pediatrics* 2002;109(4). URL: <http://www.pediatrics.org/cgi/content/full/109/4/e69>; *sleep apnea, obstructive, infant, child, tonsillectomy, meta-analysis, polysomnography, sleep disorders, snoring.*

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ABBREVIATIONS. OSAS, obstructive sleep apnea syndrome; PSG, polysomnography; ADHD, attention-deficit/hyperactivity disorder; PS, primary snoring; RDI, respiratory disturbance index; CI, confidence interval; AHI, apnea-hypopnea index; IGF, insulin-like growth factor; CPAP, continuous positive airway pressure; PPV, positive predictive value; NPV, negative predictive value; AI, apnea index.

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### INTRODUCTION

This technical report describes in detail the procedures involved in developing recommendations as given in the accompanying practice guideline on obstructive sleep apnea syndrome (OSAS).<sup>1</sup> A description of the process, methods of data compilation and analysis, and summaries of the conclusions of the committee will be given.

### FORMULATION AND ARTICULATION OF THE QUESTION ADDRESSED BY THE COMMITTEE

#### Target Audience

The practice guideline is primarily aimed at office-based pediatricians and other primary care clini-

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cians who treat children (family physicians, nurse practitioners, physician assistants). The secondary audience for the guideline includes pediatric pulmonologists, neurologists, otolaryngologists, and developmental/behavioral pediatricians.

### Definitions

The primary focus of the committee was on OSAS in childhood. The committee agreed to use the definition provided in a statement from the American Thoracic Society<sup>2</sup> with some additional elaboration of associated symptoms:

OSAS in children is a disorder of breathing during sleep characterized by prolonged partial upper airway obstruction and/or intermittent complete obstruction (obstructive apnea) that disrupts normal ventilation during sleep and normal sleep patterns. It is associated with symptoms including habitual (nightly) snoring, sleep difficulties, and/or daytime neurobehavioral problems. Complications may include growth abnormalities, neurologic disorders, and cor pulmonale, especially in severe cases. Various risk factors have been identified and are defined.

The committee sought to focus on otherwise healthy children who might have adenotonsillar hypertrophy or obesity as underlying risk factors and to specifically exclude infants younger than 1 year, children with central hypoventilation syndromes, and children at risk because of underlying abnormalities, such as craniofacial disorders; Down syndrome; cerebral palsy; neuromuscular disorders; chronic lung disease; sickle cell disease; genetic, metabolic, and storage diseases; and laryngomalacia.

### Goals of the Committee

The committee sought to address several specific goals and questions:

1. *To enhance the primary care clinician's ability to recognize OSAS.* The committee believed that a certain amount of consciousness raising would be appropriate to alert the clinician to suspect the presence of OSAS. Thus, a catalog of associated signs and symptoms is provided in the accompanying practice guideline<sup>1</sup> but will not be addressed in this technical report.
2. *To identify the most appropriate procedure for diagnosis of OSAS.* Approaches evaluated by the committee included history and physical examination, questionnaires, audiotaping or videotaping, nocturnal pulse oximetry, nap polysomnography (PSG), and ambulatory PSG, all of which would be compared with the gold standard, comprehensive overnight PSG, as defined by the American Thoracic Society.<sup>2,3</sup> In view of the fact that overnight PSG is not readily available to children in all geographic areas, consideration was given to alternative diagnostic approaches even if their accuracy is suboptimal.
3. *To identify risks associated with pediatric OSAS.* In adults, OSAS is associated with excessive daytime sleepiness (leading to cognitive defects and increased mortality attributable to susceptibility to motor vehicle crashes), pulmonary hypertension, and systemic hypertension. The committee wished

to evaluate the strength of pediatric data in this area.

4. *To evaluate management options for OSAS.* The committee sought to find data relating to adenotonsillectomy and alternative treatment modalities in the management of OSAS. The committee set out to evaluate the risk of complications after adenotonsillectomy in children with OSAS, especially given the fact that patients not suspected to have OSAS might undergo adenotonsillectomy for other indications. The postoperative complication of particular concern was respiratory compromise. Other complications of surgery, such as bleeding and pain, were not specifically addressed in relation to OSAS. In addition, data on postoperative recurrence and persistence of OSAS was sought.

## METHODS

### Literature Search

A literature search of the National Library of Medicine's PubMed database (<http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?db=PubMed>) for the years 1966–1999 was conducted in August 1999 by staff at the American Academy of Pediatrics. The search was limited to English language literature concerning children older than 2 and younger than 18 years. The following search terms were used: sleep apnea syndrome; apnea; sleep disorders; snoring; polysomnography; airway obstruction; adenoidectomy; tonsillectomy (adverse effects, mortality); and sleep-disordered breathing.mp. This search was updated in November 2000 before preparation of this technical report.

### Article Review

Titles and abstracts (when available) of articles found by the literature search were reviewed by committee members, and all those considered to be possibly relevant were marked for detailed review. Recent review articles were included to compare their bibliographies with the result of the automated literature search. Articles deemed possibly relevant were then printed and distributed to committee members for more detailed review. A literature review form was developed for this project to standardize this part of the process (Appendix 1). Because there was a large number of articles requiring evaluation, some committee members recruited residents and fellows to assist in the performance of these reviews under their supervision. Although it became clear at this point that the number of articles that could be considered high quality by conventional epidemiologic standards<sup>4–6</sup> was small, a low threshold was used to allow inclusion of any possibly relevant articles into the next level of review. At this point, articles were compiled and divided by the committee chair, additional articles were obtained by a review of literature, committee members' files were added, and committee members were assigned specific topics (as discussed previously in "Goals of the Committee") for detailed review and compilation of evidence tables. The findings of committee members were then presented at a follow-up meeting of the entire committee. A final review and compilation into evidence tables was performed by the lead author of this technical report (M.S.S.).

Calculation of prevalence, diagnostic test characteristics, and odds ratios were performed independently of the authors' reports, using data provided in the original articles. In 2 cases, authors were contacted for clarification of data. Where applicable, odds ratios from different studies were combined, using Mantel-Haenszel weights in stratified tables. Tests for heterogeneity are reported. All statistical calculations were performed using Stata 5.0 software (Stata Corporation, College Station, TX).

## RESULTS

The literature search identified 2067 articles for initial review. Titles and abstracts (when available) of these articles were divided among the 7 committee

members for perusal as an initial screening, and of those, 278 (16.2%) were retained for more detailed scrutiny. An additional 48 relevant publications were found outside this initial review. Articles were read in full if they appeared to have any relevance to childhood OSAS; included in this group were 70 general reviews and case reports or descriptive case series, which were used to allow committee members to gain a general sense of the literature and access their bibliographies. Among various committee members, the percentage of articles chosen for more detailed review ranged from 12.7% to 23.4%. This variability was statistically significant ( $P = .007$  by Pearson  $\chi^2$ ). Excluding the methodologist, whose approach was the most permissive (23.4% acceptance rate), the range was 12.7% to 19.4%, and variability was not statistically significant ( $P = .143$  by Pearson  $\chi^2$ ) among remaining committee members.

A total of 113 articles were found that contained original data relevant to the specific aims of this committee. Most of the publications that were reviewed in detail provided little quantitative data for analysis. In particular, most papers that were older than 5 years presented case series or poor-quality cohort studies or omitted important details. These lower-quality studies provide the background for current expert opinion but are otherwise of limited value and will not be listed in detail for this report. Studies that were of quantitative value are tabulated and were given quality ratings<sup>4,6</sup> in the tables. Briefly, rating levels of studies on treatment efficacy were assigned as follows<sup>4</sup>:

Level I—Randomized trials with low rates of false-positive ( $\alpha$ ) and/or false-negative ( $\beta$ ) results (high power).

Level II—Randomized trials with high rates of false-positive ( $\alpha$ ) and/or false-negative ( $\beta$ ) results (low power).

Level III—Nonrandomized concurrent cohort comparisons between contemporaneous patients who did and did not receive an intervention, or case-control or cross-sectional studies with appropriate control group.

Level IV—Nonrandomized historical cohort comparisons between current patients who received an intervention and former patients (from the same institution or from the literature) who did not, or case-control or cross-sectional studies for which control groups were suboptimally chosen.

Level V—Case series without controls.

Rating levels for diagnostic tests were assigned as follows<sup>6</sup>:

Level 1—Independent blind comparison of patients from an appropriate spectrum of patients, all of whom have undergone both the diagnostic test and the reference standard.

Level 2—Independent blind or objective comparison performed in a set of nonconsecutive patients or confined to a narrow spectrum of study individuals (or both), all of whom have undergone both the diagnostic test and the reference standard.

Level 3—Independent blind or objective comparison of an appropriate spectrum of patients, but the reference standard was not applied to all.

Level 4—Reference standard was unobjective, unblinded, or not independent; positive and negative tests were verified using separate reference standards; or study was performed in an inappropriate spectrum of patients.

### Prevalence of Snoring and OSAS

We found 7 studies that attempted to establish prevalence of snoring in childhood.<sup>7–13</sup> These studies came from a variety of European countries, and all ascertained data via parent questionnaire. The prevalence of snoring in these studies ranged from 3.2% to 12.1%, which was significantly heterogeneous ( $P < .0001$ ). The study by Gislason and Benediksdottir<sup>8</sup> seemed to be somewhat of an outlier, especially because the frequency of OSAS they reported was nearly the same as that of snoring, but with omission of that study, the heterogeneity remains significant ( $P < .001$ ). Characteristics of the studies are shown in Table 1, and prevalence estimates are seen in Fig 1.

Three studies reported on prevalence of OSAS, and estimates ranged from 0.7% to 10.3%. All used very different criteria, including 1 that gave estimates based on 2 different criteria.<sup>14</sup> The variability of these estimates was so great that no attempt was made to combine the data, which are summarized in Table 1.

### Sequelae of OSAS

Most published articles on complications of OSAS are reports of retrospective case series or prospectively collected, uncontrolled data comparing measures before and after surgical treatment. These articles are summarized in this report and in evidence tables (Tables 2–4).

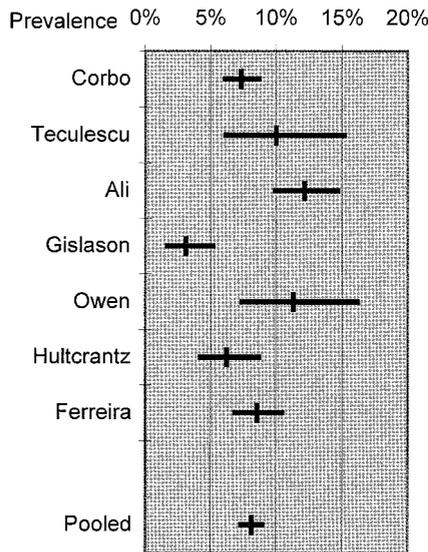
### Cognitive and Behavioral Abnormalities

The committee found 12 publications that evaluated the association of behavioral problems, especially hyperactivity or attention-deficit/hyperactivity disorder (ADHD), with sleep-disordered breathing (Table 2). In an early case series<sup>15</sup> of 50 children with OSAS documented by PSG, 84% had excessive daytime sleepiness, 76% had some behavior disturbance, 42% were hyperactive, and 16% had decreased school performance. A number of cross-sectional studies have been done that compare the risk of behavioral problems in children who snore with that of a control population. None of these studies distinguish children with OSAS from those with primary snoring (PS). For the study of Weissbluth et al,<sup>16</sup> parents of children attending a general pediatric practice were surveyed, and 71 children were reported to have behavioral or academic problems. Snoring, mouth breathing, and labored breathing when asleep were reported to be more than twice as common in these children as in the comparison group. The study of Chervin et al<sup>17</sup> reported that 33% of children with ADHD were habitual snorers, compared with 11% of children attending a general psy-

**TABLE 1.** Prevalence of Snoring and OSAS

Author	Year	Number	Country	Age (Years)	Frequency of Snoring		
					Habitual	Sometimes	Never
Prevalence of Snoring							
Corbo <sup>12</sup>	1989	1615	Italy	6–13	7.3%	8.5%	50.9%
Teculescu <sup>11</sup>	1992	190	France	5–6	10%	22.1%	54.2%
Ali <sup>7</sup>	1993	782	United Kingdom	4–5	12.1%	ND	ND
Gislason <sup>8</sup>	1995	454	Iceland	0.5–6	3.2%	16.7%	ND
Owen <sup>9</sup>	1995	222	United Kingdom	1–10	11%	ND	ND
Hultcrantz <sup>10</sup>	1995	500	Sweden	4	6.2%	18%	ND
Ferreira <sup>13</sup>	2000	1381	Portugal	6–11	8.6%	30.6%	60.1%
Prevalence of OSAS							
Ali <sup>7</sup>	1993	782 screened; 132 monitored	United Kingdom	4–5	Pulse oximetry, video	0.7%	
Gislason <sup>8</sup>	1995	454	Iceland	0.5–6	PSG, AHI >3	2.9%	
Redline <sup>14</sup>	1999	126	United States	2–18	Home PSG	1.6% (AHI>10) 10.3% (AHI>5)	

ND indicates no data provided; PSG, polysomnography.



**Fig 1.** Point estimates of the prevalence of snoring in the general pediatric population, as per various publications, with 95% CI and the pooled estimate.

chiatric clinic and 9% attending a general pediatric clinic. A previously mentioned prevalence study by Ali et al<sup>7</sup> found that children who were reported to snore during most nights were also reported to have more daytime sleepiness and hyperactivity than were children in a comparison group. Children were evaluated using Conners scales, and those with more severe sleep disturbance were also more likely to be at the 95th percentile on Conners subscales relating to hyperactive, inattentive, and aggressive behaviors. In a follow-up report,<sup>18</sup> 29 of 60 children reported to snore 2 years previously no longer snored (weighted  $\kappa$  0.52), although habitual snoring was again found to be associated with daytime sleepiness and hyperactivity. In another publication, the same authors identified 12 children with sleep-disordered breathing (by overnight pulse oximetry and videotaping), 11

snoring children without sleep-disordered breathing, and a control group not undergoing adenotonsillectomy and administered Conners scales, the Continuous Performance Test (a test of attention), and the Matching Familiar Figures Test (a test of vigilance).<sup>19</sup> Significant improvement was found in Conners parent scale scores for aggressive, inattentive, and hyperactive behaviors; attention; and vigilance in the sleep-disordered breathing group after adenotonsillectomy, and similar changes were found in the primary snorers postoperatively, but not in control groups. In a recent population-based cross-sectional study of 988 Portuguese children, Ferreira et al<sup>13</sup> found that habitual snorers were twice as likely as nonsnorers to have an abnormal score on the Children's Behavioral Questionnaire. Finally, Blunden et al<sup>20</sup> compared 16 children referred for adenotonsillectomy or snoring with a control group and found impaired selective and sustained attention scores only in the children who snored. They also reported that the snoring children had significantly lower average IQ scores. PSG was done in these children, but for purposes of analysis, the PS and OSAS groups were combined because preliminary analyses revealed no significant group differences on any neurocognitive or behavioral parameter (all  $P > .05$ ). The authors pointed out that even patients with OSAS had very mild abnormalities (mean respiratory disturbance index [RDI] <1). Although it is possible that lack of power and selection bias may have contributed to their findings, the implication is that snoring without overt OSAS might be associated with neurocognitive abnormalities.

Although the 6 cross-sectional studies described previously all reported on slightly different behavioral and cognitive phenomena, their findings were pooled, and the results of this are shown in Fig 2. The Mantel-Haenszel test for heterogeneity was not significant ( $P = .4577$ ). The combined odds ratio for

**TABLE 2.** Association of Behavioral Abnormalities and OSAS

Author	Year	Number	Age	Diagnostic Technique	Study Methodology and Rating	Results and Comments
Guilleminault <sup>15</sup>	1981	50	NS	PSG	Case series, level V	Of children with OSAS: 84% had excessive daytime sleepiness; 76% had some behavior disturbance; 42% were hyperactive; 16% had decreased school performance.
Weissbluth <sup>16</sup>	1983	71 with behavior problems; 355 controls	74 ± 5 mo	Questionnaire	Cross-sectional, level III	Of children with behavior problems: OR <sub>snoring</sub> : 2.3 (1.3, 4.1); OR <sub>abored breathing</sub> : 2.6 (1.1, 6.1).
Ali <sup>7</sup>	1993	782	4-5 y	Questionnaire	Cross sectional, level III	For habitual snorers: OR <sub>daytime sleepiness</sub> : 1.2 (1.0, 1.4); OR <sub>hyperactivity</sub> : 1.7 (1.4, 2.2). Connors subscales (parents): OR <sub>aggressive behavior</sub> : 9.3 (2.3, 39.1); OR <sub>inattentive behavior</sub> : 5.1 (1.2, 21.8); OR <sub>hyperactive behavior</sub> : 9.5 (2.3, 39.6).
Ali <sup>18</sup>	1994	507	6-7 y	Questionnaire	Cohort (f/u of 1993 study); cross-sectional, level III	For habitual snorers: RR <sub>hyperactivity</sub> : 2.8 (1.6, 4.7); RR <sub>daytime sleepiness</sub> : 6.1 (2.5, 14.9).
Ali <sup>19</sup>	1996	12 with OSAS, 11 with PS, 10 controls	>6 y	Overnight pulse oximetry, videotape	Cohort, level III	29 of 60 habitual snorers were still snoring at follow-up. Aggression, inattention, hyperactivity, and vigilance improved postoperatively in the OSAS and PS groups, not in the controls.
Chervin <sup>17</sup>	1997	143	2-18 y	Questionnaire	Cross-sectional, level III	Habitual snoring was found in: 33% of ADHD patients (OR = 4.7 [1.5, 14.7]); 11% of general psychiatry patients (OR = 1.1 [0.3, 4.0]); and 9% of general pediatrics patients (OR = 1.0).
Gozal <sup>23</sup>	1998	297 screened, 54 with abnormal sleep	First grade	Questionnaire, home overnight pulse oximetry	Cohort, level III	18.1% of children in lowest 10th percentile academically had gas exchange abnormalities during sleep. Mean school grades improved in 24 children who received adenotonsillectomy, but not in 30 who did not ( <i>P</i> < .001).
Harvey <sup>24</sup>	1999	24 postoperative adenotonsillectomy patients, 15 untreated	NS	Overnight PSG	Cohort, level IV	Children who had adenotonsillectomy showed no improvement in behavioral measures.
Rosen <sup>22</sup>	1999	326 total, 192 with OSAS	1-12 y	Overnight PSG	Case series, level V	Of habitual snorers (OSAS = non-OSAS): 19% had daily tiredness; 10% had excessive daytime sleepiness; 9% had behavior, school, or mood problems.
Goldstein <sup>21</sup>	2000	36 preoperative, 15 postoperative	2-18 y	Clinical evaluation	Case series, level V	Of children with clinically diagnosed sleep apnea: 28% had abnormal Child Behavior Checklist scores preoperatively; 13% had abnormal scores postoperatively ( <i>P</i> = NS). Mean score was significantly lower postoperatively ( <i>P</i> < .001).
Ferreira <sup>13</sup>	2000	84 snorers, 593 nonsnorers	6-11 y	Questionnaire	Cross-sectional, level III	24% of habitual snorers had abnormal Children's Behavior Questionnaire score, compared with 15% of nonsnorers (OR = 1.8 [1.0, 3.3]).
Blunden <sup>20</sup>	2000	16 snorers, 16 controls	5-10 y	Overnight PSG (but OSAS and PS combined)	Cross-sectional, level III	Mean WISC-III IQ was 12.2 points lower in snorers ( <i>P</i> < .01). Six of 16 snorers and 0 of 16 controls had impaired selective attention scores; 3 of 16 snorers and 0 of 16 controls had impaired sustained attention scores.

NS indicates not specified; PSG, polysomnography; OR, odds ratio; f/u, follow-up; RR, relative risk; WISC-III IQ, Wechsler Intelligence Scale for Children—Third Edition.

**TABLE 3.** Association of Growth Abnormalities and OSAS

Author	Year	Number	Age (Years)	Diagnostic Technique	Study Methodology and Rating	Results and Comments
Lind <sup>25</sup>	1982	14 with obstructed breathing, 6 controls	5	Capnography, clinical evaluation	Case series, level V	Growth improved after tonsillectomy.
Williams <sup>26</sup>	1991	37	0–3	Clinical evaluation	Case series, level V	Effect of adenotonsillectomy on median weight percentile: preoperatively, 5; postoperatively, 35 ( $P < .0001$ ). Median height percentile: preoperatively, 5; postoperatively, 23 ( $P = .1$ ).
Marcus <sup>27</sup>	1994	14	2–6	PSG: AI >1	Case series, level V	Effect of adenotonsillectomy on weight z score: preoperatively, $-0.30$ ; postoperatively, $0.04$ ( $P < .005$ ). Caloric intake unaltered, energy expenditure decreased.
Bar <sup>28</sup>	1999	13	1–11	PSG (10) or overnight oximetry (3)	Case series, level V	Effect of adenotonsillectomy on weight z score: preoperatively, $0.9$ ; postoperatively, $1.2$ ( $P < .01$ ). Height z score: preoperatively, $0.7$ ; postoperatively, $0.8$ ( $P = NS$ ). Serum IGF-1 levels increased postoperatively.
Soultan <sup>29</sup>	1999	45 (17 obese)			Cohort, level III	Effect of adenotonsillectomy on weight z score: preoperatively, $1.4$ ; postoperatively, $2.0$ ( $P < .001$ ). Height z score: preoperatively, $0.0$ ; postoperatively, $0.6$ ( $P < .01$ ). 69% had weight gain postoperatively (59% of obese patients).

PSG indicates polysomnography; NS, not significant.

neurobehavioral abnormalities in snoring children is 2.93 (95% confidence interval [CI], 2.23–3.83).

Several other papers are of interest but cannot be quantitatively combined. Goldstein et al<sup>21</sup> had parents of 36 children who were referred for adenotonsillectomy because of clinically significant obstructive symptoms complete the Child Behavior Checklist and found that 10 (28%) had abnormal results. Postoperatively, 15 had a repeat evaluation, and only 2 (13%) still had abnormal results. This finding was not clinically significant, presumably because of lack of statistical power, but there was a clinically significant improvement in mean test scores postoperatively ( $P < .001$ ).

Four studies used overnight PSG testing to establish a diagnosis of OSAS. Rosen<sup>22</sup> reported on a series of 326 children referred for evaluation of snoring, of whom 59% met PSG criteria for OSAS. Daily tiredness was reported in 19%; excessive daytime sleepiness was reported in 10%; and behavior, school, or mood problems were reported in 9%, with no difference between the OSAS and non-OSAS groups. Gozal<sup>23</sup> conducted an interesting study in which 297 first graders who were in the lowest 10th percentile academically were evaluated for sleep-disordered breathing by parent questionnaire combined with overnight (home) oximetry. Adenotonsillectomy was recommended in the 54 children (18.1%) with abnormal test results; 24 accepted surgery, and 30 did not. Mean grades increased from  $2.43 \pm 0.17$  to  $2.87 \pm 0.19$  in the children who had surgery, with no change in the untreated OSAS group or the non-OSAS group ( $P < .001$ ).

The findings of an Australian study<sup>24</sup> contrasted with the aforementioned papers. Thirty-nine children with PSG evidence of OSAS were followed 6 months after the initial PSG, and 24 had received adenotonsillectomy. These were compared with chil-

dren who were waiting for intervention ( $n = 5$ ; median apnea-hypopnea index [AHI] = 5.5) or didn't require intervention ( $n = 10$ ; median AHI = 3.1). Information on AHI was not given for the surgical group. At follow-up, children in the surgical and nonsurgical groups had improved sleep behavior. Intervention did not result in any statistically significant improvement in development or temperament, although the study was probably underpowered.

In summary, studies generally show a nearly threefold increase in behavior and neurocognitive abnormalities in children with sleep-disordered breathing. Most of these studies did not definitively differentiate children with PS from those with OSAS, so the true prevalence of behavior and learning problems in children with OSAS versus PS is not clear. It is possible, however, that PS, even in the absence of clear-cut OSAS, might place children at risk.

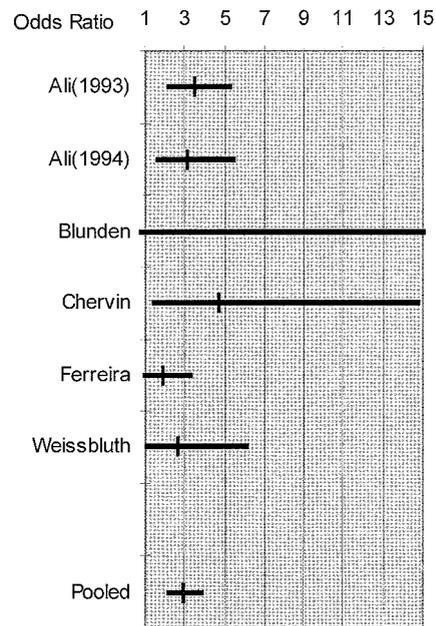
#### Growth

Four studies evaluating growth and OSAS were found. Marcus et al<sup>27</sup> evaluated 14 prepubertal children with a mean age of 4 years  $\pm$  1 standard deviation who had OSAS documented by overnight PSG and measured caloric intake and sleeping energy expenditure as well as anthropomorphic measurements before and after adenotonsillectomy. Average sleeping energy expenditure decreased, and mean weight z score increased postoperatively without any change in caloric intake. Bar et al<sup>28</sup> evaluated changes in growth and also measured insulin-like growth factor (IGF)-I and IGF-binding protein-3 levels before and 18 months after adenotonsillectomy. Both studies showed statistically significant increases in weight but not height; IGF-I levels increased and IGF-binding protein levels did not. An interesting report on the effect of adenotonsillectomy on growth included a group of obese and morbidly

**TABLE 4.** Association of Cardiovascular Complications and OSAS

Author	Year	Number	Age (Years)	Diagnostic Technique	Study Methodology and Rating	Results and Comments
Laurikainen (Finland) <sup>30</sup>	1992	19 for adenotonsillectomy; 14 with OSAS	3-7	PSG with static charge sensitive bed	Case series, level V	Electrocardiographic evidence of RVH in 21% with AI >5 and 20% with AI <5. RVH resolved after adenotonsillectomy.
Wilkinson (United Kingdom) <sup>31</sup>	1981	92 for adenotonsillectomy	1-13	Clinically determined need for adenotonsillectomy	Case series, level V	3% of children had right heart strain on electrocardiography, reversed with adenotonsillectomy.
Shiomi (United States) <sup>32</sup>	1993	6 with OSAS	3-14	PSG, AHI >3	Case series, level V	50% showed leftward shift of interventricular septum by echocardiography correlating with P <sub>es</sub> .
Tal (Israel) <sup>33</sup>	1988	27 with clinical OSAS	0.75-7.5	Questionnaire by Brouillette et al, OSAS score >3.5	Case series, level V	RVH in 7% by echocardiography; RV ejection fraction reduced in 37%; all 11 with postoperative f/u were improved.
Marcus (United States) <sup>34</sup>	1998	41 with OSAS, 26 with PS	1-18	PSG, AI >1	Cross sectional, level III	Compared with children with PS, children with OSAS had higher diastolic (but not systolic) pressure.

PSG indicates polysomnography; RVH, right ventricular hypertrophy; P<sub>es</sub>, esophageal pressure; RV, right ventricle; f/u, follow-up.



**Fig 2.** Point estimates of the odds ratio for hyperactive behavior in children with habitual snoring, as per various publications, with 95% CI and the pooled estimate.

obese children<sup>29</sup> and documented postoperative increases in weight and height, even in those children who were initially obese. The other 2 studies,<sup>25,26</sup> which had poorer documentation of OSAS, reported similar results. None of these studies reported a comparison with a nonsurgical control group, comparison with children operated on for indications other than OSAS, or comparison with children who had PS.

#### Cardiovascular

Eight case reports or small series were found that documented cor pulmonale or hypertension, which reversed with adenotonsillectomy or other surgical correction in patients with clinically diagnosed OSAS.<sup>35-42</sup> Two case series<sup>30,31</sup> reported children with adenotonsillar hypertrophy with or without clinical airway obstruction who were found to have right ventricular dysfunction that reversed after adenotonsillectomy. One study<sup>32</sup> described pulsus paradoxus and leftward shift of the interventricular septum secondary to snoring in 3 of 6 children with OSAS. This correlated with negative esophageal pressures but not with oxygen desaturation, and it reversed with nasal continuous positive airway pressure (CPAP). Tal et al<sup>33</sup> used radionuclide ventriculography to evaluate ventricular function in 27 children referred for oropharyngeal obstruction who had abnormal Brouillette questionnaire scores for OSAS; PSG was not performed. They found decreased right ventricular ejection fraction in 37% of these children and abnormal wall motion in 67%. All of the 11 patients who had a repeat evaluation after adenotonsillectomy showed improvement. Systemic blood pressure was evaluated in a study of children referred for PSG.<sup>34</sup> Higher diastolic pressures (adjusted for body mass index and age) were found in children with OSAS, compared with those with PS.

The prevalence of blood pressure measurements >95th percentile was high in both groups (32% vs 19%, respectively), with a nonsignificant difference that may have been attributable to low power. The response after adenotonsillectomy was not reported. This was the only study that compared cardiovascular complications in children with OSAS versus those with PS.

#### Miscellaneous

One study<sup>43</sup> reported on 115 enuretic children undergoing adenotonsillectomy for any indication. There was a 66% reduction in enuretic nights 1 month after surgery and a 77% decrease 6 months after surgery. In the group with secondary enuresis, 100% were dry 6 months after surgery.

### Diagnosis of OSAS

#### Polysomnography

One of the problems in evaluating various methods of diagnosing OSAS in children is that the gold standard, overnight PSG, has not been well standardized in its performance or interpretation. Although recent consensus statements pertaining to standards and normative data<sup>2,3</sup> should lessen this problem, the question of definition remains problematic. Pediatric sleep specialists use the adult model in describing a continuum of sleep-disordered breathing from PS to upper airway resistance syndrome to obstructive hypoventilation and OSAS. It is assumed that PS is a benign condition and OSAS is associated with undesirable complications. Normative standards for their polysomnographic determination have been chosen on the basis of statistical distribution of data,<sup>44,45</sup> but it has not been established that those standards have any validity as predictors of the occurrence of complications. In other words:

“On the basis of normative data, an obstructive apnea index of 1 is often chosen as the cutoff for normality. However, while an apnea index of 1 is statistically significant (ie, at the 97.5th percentile for an asymptomatic, normative population), it is not known what level is clinically significant.”<sup>3</sup>

Of the few studies that compare children with polysomnographically defined OSAS with those with PS in regard to prevalence of complications,<sup>19,22,30,34</sup> only 1 found a clear difference between the 2 groups.<sup>34</sup> This is an important point, because with a poorly validated gold standard, statements regarding diagnostic accuracy of alternative methods of diagnosis become dubious.<sup>46</sup> Finally, the test-retest reliability of overnight PSG, which in adults is no greater than 91% and possibly somewhat lower,<sup>47</sup> has never been evaluated in children.

Having stated these points, additional analysis of the validity of alternative diagnostic approaches will be done assuming PSG as the gold standard. One additional benefit of overnight PSG is that in addition to establishing the diagnosis of OSAS, PSG also may be used to determine its severity. It has been suggested that the severity of OSAS is an important predictor of complications, particularly in the immediate postoperative period.<sup>48</sup> None of the alternative

diagnostic techniques discussed below have been evaluated for this purpose.

#### Questionnaires

In 1984, Brouillette et al<sup>49</sup> reported high accuracy for a diagnostic questionnaire for OSAS in children with adenotonsillar hypertrophy. This questionnaire was initially tested on 23 children with OSAS and 46 controls. On the basis of this questionnaire, a 3-variable discriminant function was calculated as follows:

$$\text{OSAS score} = 1.42D + 1.41A + 0.71S - 3.83$$

where D is difficulty during sleep, A is apnea observed during sleep, and S is snoring. Values assigned to D and S were: 0 = never; 1 = occasionally; 2 = frequently; and 3 = always. Values assigned to A were: 0 = no; and 1 = yes. This system was then applied to a prospective group of 23 patients referred for evaluation of possible OSAS. The authors demonstrated that a score of >3.5 perfectly predicted the presence of OSAS by PSG; a score of <-1 perfectly predicted absence of OSAS; and a score in between was indeterminate. Unfortunately, there were 5 children who were believed to have a borderline PSG, confusing the issue somewhat. It appears that the choices of 3.5 and -1 as breakpoints in the score were made posthoc and were, thus, somewhat arbitrary. Since the initial publication of the questionnaire, 3 additional studies have been published detailing the results of its use. The results of these are provided in Table 5. All of these studies prospectively evaluated similar groups of pediatric patients with a similar prevalence of OSAS; PSG with similar evaluation criteria was performed on all subjects, and all completed the same questionnaire applied in similar ways. This scoring system is sufficiently simple and straightforward, so its application can be expected to be fairly standard and replicable. Thus, data from these studies was combined, and conclusions were drawn accordingly.

As can be seen from Table 5, the OSAS questionnaire by Brouillette et al performed much less well in subsequent applications. The 4 studies (including a later study by the same authors) included a total of 765 patients with an overall prevalence of OSAS confirmed by PSG of 60%. Applied to these patients, the score was indeterminate in 47%; in subjects who were categorized (ie, not indeterminate), its positive predictive value (PPV) was 65% and negative predictive value (NPV) was 46%. Using the pooled data for calculation, the likelihood ratio of positive questionnaire results is 1.24, and the likelihood ratio of negative questionnaire results is 0.78. Overall, the use of the questionnaire by Brouillette et al as a substitute for PSG would clearly be fraught with error, leading to numerous false-positive and false-negative results in the diagnosis of OSAS.

Other publications reporting attempts at creating questionnaires or developing other purely clinical criteria to substitute for PSG are uninterpretable because of their failure to compare their criteria with PSG<sup>50-53</sup> or unsuccessful in developing any re-

**TABLE 5.** Studies Using Questionnaires or Clinical Criteria

Author	Year	Methodology Rating	PSG Criterion for OSAS	Number	Prevalence of OSAS	Indeterminate*	Se	Sp	PPV	NPV
Studies Using Questionnaire by Brouillette et al										
Brouillette et al <sup>49</sup>	1984	2	Pediatric criteria without AHI	21†	67%	71%	100%	100%	100%	100%
Carroll <sup>58</sup>	1995	1	OSAS = AHI ≥1	70	70%	50%	78.6%	71.4%	64.7%	83.3%
Rosen <sup>22‡</sup>	1999	1	OSAS = AI ≥1	325	50%	50%	83.3%	58.5%	76.9%	67.9%
Brouillette et al <sup>59</sup>	2000	1	OSAS = AHI ≥1	349	60%	44%	34.7%	39.2%	48.3%	26.9%
Total of above 4 studies				765	59%	47%	59.5%	51.9%	65.3%	45.7%
Studies Using Other Questionnaires or Clinical Criteria										
van Someren <sup>51§</sup>	1990	3	Pulse oximetry, no PSG	44	34%		93.3%	82.8%	73.7%	96.0%
van Someren <sup>52  </sup>	2000	3	Video/pulse oximetry, no PSG	120	46%		59.1%	72.5%	60.5%	71.4%
Niemenen <sup>55</sup>	1997	2	OSAS = AHI ≥1	78	37%	RRs of individual items on questionnaire were calculated (highest was 3.6); data on sensitivity, specificity not given.				
Leach <sup>54</sup>	1992	1	Pediatric criteria without AHI	93	37%	RRs of individual symptoms were calculable but none were striking; data on sensitivity, specificity not given.				
Wang <sup>57</sup>	1998	1	OSAS = AHI ≥5	82	30%	Highest PPV for various clinical parameters was 46% for enuresis; highest NPV was 100% for small tonsils.				
Suen <sup>48</sup>	1995	1	OSAS = AHI >5	69	51%	"No significant correlations were noted between RDI and any of the clinical variables . . ."; specific data not given.				
Croft <sup>53</sup>	1990	3	Pulse oximetry, observation	50	26%	Snoring and apnea by report and observation correlated poorly; mild correlation between sleep obstruction and both tonsillar position and lateral cephalometry, but not nasal airway.				
Silvestri <sup>50</sup>	1993	3	Abbreviated PSG, without AHI	32	66%	A retrospectively created score combining presence of ≥5 symptoms, adenotonsillar enlargement, IBW ≥200% was accurate in 81%; specific data not given.				

Se indicates sensitivity; Sp, specificity; PSG, polysomnography; RR, relative risk; IBW, ideal body weight.

\* Percentage of patients whose score fell into the indeterminate range.

† Twenty-three subjects were evaluated, but data was reported on only 21.

‡ Unpublished details pertaining to the data were obtained by personal communication with the primary author.

§ Clinical criteria of mouth breathing, audible respiration at rest, and an awake arterial oxygen saturation of <96%, compared with results of pulse oximetry.

|| Referring clinician's impression of moderate to severe sleep-related upper airway obstruction compared with results of video/pulse oximetry system.

liable predictive criteria.<sup>48,54-57</sup> They are tabulated in Table 5.

#### Audiotaping and Videotaping

Two studies have evaluated the use of home audiotaping, and 1 evaluated home videotaping, as a screening test for OSAS. The methods used to evaluate these techniques were different, so the data do not lend themselves to pooling. Sivan et al<sup>60</sup> scored a 30-minute videotape in 58 children using 7 variables, including loudness and type of inspiratory noise, movements during sleep, number of waking episodes, number of apneas, chest retractions, and mouth breathing. The PSG results were abnormal in 62%. They reported a sensitivity of 94%, specificity of 68%, PPV of 83%, and NPV of 88%. Posthoc analysis (similar to what was done in the 1984 study by Brouillette et al) was performed in 2 ways, leading to the development of an indeterminate score and better test characteristics in the categorizable group. As

might be expected, a scoring system that places a greater number of subjects into the indeterminate group leads to better NPV and PPV. The results of all 3 approaches are shown in Table 6.

Goldstein et al<sup>61</sup> developed a 7-item predictive score that considered the presence of snoring, respiratory pauses, gasping, sleeping with neck extended, daytime sleepiness, adenoid facies, and the presence of pauses in breathing of at least 5 seconds on an audiotape recorded by the parents. The criteria used to score each category were not precisely described, and there seemed to be significant variability in the evaluation of the audiotapes, which were reviewed for "at least 2 minutes . . . for each child, and an average of 10 minutes were generally reviewed. Various parts of the tape were sampled." Patients were categorized as definitely, possibly, or not likely having OSAS on the basis of these items, but no description was provided of how these items were scored and combined, and no measure of interobserver vari-

**TABLE 6.** Studies Comparing Alternative Testing Procedures With Overnight PSG

Author	Year	Methodology Rating	PSG Criterion for OSAS	Number	Prevalence	Indeterminate*	Se	Sp	PPV	NPV
I. Audio/Videotaping Sivan <sup>60</sup>	1996	1	OSAS = AHI ≥1	58	62%	45%	94.4%	68.2%	82.9%	88.2%
							100%	100%	100%	100%
							16%	88.9%	77.3%	86.5%
Goldstein <sup>61</sup>	1994	2	OSAS = AHI ≥15	30	43%		92.3%	29.4%	50.0%	83.3%
Lamm <sup>64†</sup>	1999	1	OSAS = AHI ≥5	29	48%		71%	80%	75%	73%
II. Pulse Oximetry Brouillette et al <sup>59</sup>	2000	3	OSAS = AHI ≥1	349	60%	46%	42.9%	97.8%	96.8%	53.1%
III. Nap PSG Marcus <sup>65</sup>	1992	4	Descriptive	40	95%		73.7%	100%	100%	16.7%
Saeed <sup>66</sup>	2000	3	OSAS = AHI ≥1	143	66%		68.4%	60.4%	77.4%	49.2%

PGS indicates polysomnography; Se, sensitivity; Sp, specificity.

\* Percentage of patients whose score fell into the indeterminate range.

† Test characteristics reported as the mean for 7 separate observers.

ability was attempted, so reproducibility is unknown. A total of 30 children were studied prospectively, of whom 13 (43%) had OSAS confirmed by PSG. The authors reported a sensitivity of 92.3%, specificity of 29.4%, PPV of 50.0%, and NPV of 83.3%, which they calculated by combining the “definite” and “possible” groups into a positive screening category. If their possible group was eliminated from consideration (analogous to the way Brouillette et al treated indeterminate scores<sup>49</sup>), a mild decrease in sensitivity (91%) and mild increases in specificity (38%) and PPV (56%) are seen.

Goldstein et al concluded that children whose results of evaluation for sleep apnea (as performed using their technique, including audiotaping) are negative do not need PSG, because the sensitivity of their clinical assessment is high. They recommended PSG for children who appear to have OSAS, because the specificity of clinical assessment is low. It is important to note that the percentage of positive results of PSG in their study (43%) was somewhat lower than the prevalence of approximately 60% reported in most studies of children referred for evaluation of possible OSAS. This is probably (at least in part) because they used more restrictive PSG criteria for diagnosing OSAS (AHI >15). If a population with a higher prevalence of OSAS were studied, it is likely that the PPV of the clinical evaluation by Goldstein et al would be higher and the NPV would be lower.<sup>a</sup> In addition, the higher AHI as a diagnostic criterion might have biased the study toward the more severe end of the OSAS spectrum. The possibility of spectrum bias<sup>62</sup> and the undocumented reproducibility<sup>63</sup> of the tape evaluation raise the question of whether test characteristics will be as good if applied to a large, general population.

<sup>a</sup> Using the authors’ calculations for sensitivity and specificity but a pretest probability of 60% rather than 43% to calculate likelihood ratios, the post-test probability of having OSAS with a negative score using the technique of Goldstein et al is 26%, higher than their paper indicates for a lower risk group. The post-test probability of a positive result in a higher prevalence group is 69% (ie, 31% rate of false positivity).

$$\begin{aligned} \text{likelihood ratio of a positive test result} &= (\text{sensitivity}) / (1 - \text{specificity}) \\ \text{likelihood ratio of a negative test result} &= (1 - \text{sensitivity}) / (\text{specificity}) \\ \text{pretest odds} &= (\text{prevalence}) / (1 - \text{prevalence}) \\ \text{post-test odds} &= \text{pretest odds} \times \text{likelihood ratio} \\ \text{post-test probability} &= (\text{post-test odds}) / (\text{post-test odds} + 1) \end{aligned}$$

A second study of the use of home audiotaping as an abbreviated test for OSAS<sup>64</sup> used 7 observers to analyze audiotapes of 29 children referred for evaluation; 48% were subsequently found to have positive PSG. Observers listened to 15 minutes of audiotape and specifically scored the presence of struggle sounds and respiratory pauses. A mean  $\kappa$  statistic of 0.70 (range, 0.50–0.93) was calculated, indicating moderately good interobserver agreement. The presence of a struggle sound on the audiotape gave the best posthoc test characteristics, with a sensitivity of 0.71, specificity of 80%, NPV of 73%, and PPV of 75%.

To summarize, the use of home audiotaping and videotaping has been inadequately investigated. Additional studies are necessary. It should be pointed out that there was no consensus of the committee regarding acceptable rates of false-negative and false-positive results for tests used as an alternative to PSG.

#### Pulse Oximetry

Seven studies were found that reported on pulse oximetry in children suspected of having OSAS.<sup>9,19,23,51,59,67,68</sup> However, only 1 compared pulse oximetry to PSG. In this study<sup>59</sup> involving 349 children, pulse oximetry was performed during PSG and was evaluated independently of the PSG interpretation, with well-defined criteria and excellent interobserver agreement. There were 89 PSGs (25.5%) performed in a sleep lab; the others were done at home, so the gold standard was not identical for all subjects. In this group, with a 60.2% prevalence of OSAS, the PPV was 97% (90 of 93). However, the NPV of the test (calculated by the authors by combining subjects with either inconclusive or negative tests) was only 53%. When the analysis was limited to subjects without any medical diagnoses other than adenotonsillar hypertrophy, the PPV was 100%, with an insubstantial change in NPV.

Given the test characteristic described, it appears that overnight pulse oximetry could provide an accurate screen for OSAS, insofar as a positive result may be a good predictor of an abnormal PSG result. However, the findings of the single study described in this report need to be replicated.

### *Nap Polysomnography*

Two papers from the same institution have evaluated the utility of brief (1 hour) daytime nap studies in comparison with full overnight PSGs (Table 6). The conclusions of both are generalizable to only a limited degree, however. Marcus et al<sup>65</sup> studied 40 children referred for evaluation of possible OSAS, but this group was not representative of the type of patient addressed by this practice guideline, because only 35% had adenotonsillar hypertrophy as the underlying cause of their sleep disturbances. Other diagnoses included Down syndrome (40% of subjects), various upper airway abnormalities, and other neurologic and respiratory problems. Furthermore, 95% (38 of 40) of the patients studied had abnormal overnight PSG results, providing little opportunity to evaluate the test performance of nap studies in children with normal PSG results. The study by Saeed et al<sup>66</sup> limited itself to children addressed by this practice guideline (age, 1–18 years; adenotonsillar hypertrophy; absence of other significant disease). They reported on the results of overnight PSG in children with normal and mildly abnormal nap study results. Patients with severely abnormal nap study results were excluded; they were assumed to have significant OSAS and, therefore, referred directly for tonsillectomy without overnight PSG (S.D. Ward, personal communication). In this group, for which prevalence of PSG-documented OSAS was 66%, the nap studies had a PPV of 77% and NPV of 49%. In fact, if children with more severely abnormal nap study results were included in the analysis and the investigators are correct in their assumption that these children all have abnormal overnight PSG results, then the sensitivity and PPV of nap studies is actually higher than that reported in this paper. Thus, it is possible that abnormal nap study results might provide a predictive value adequate to allow the recommendation for surgery without corroborative overnight PSG, but confirmation of this conclusion (asserted by the authors) is lacking. On the other hand, a nap study with negative results would still require a follow-up overnight PSG for confirmation.

### *Home Polysomnography*

One group has published data comparing the results of PSG performed in children at home with those performed at the sleep laboratory.<sup>69,70</sup> In a report of 21 children between the ages of 2 and 12 years who were studied in both environments, the sensitivity and specificity of home PSG varied depending on the severity of OSA. When an AHI >1 was used as the criterion for diagnosing OSAS, the sensitivity of home PSG was 100% and the specificity was 62%; for AHI >3, sensitivity was 88% and specificity was 77%; for AHI >5, sensitivity and specificity were both 100%. This group uses a sophisticated type of ambulatory PSG that is not commercially available and is not analogous to commercial systems. Also, their system did not allow for detection of obstructive hypoventilation. Furthermore, the subjects used in their report were not chosen sequentially or at random, and the authors describe a com-

plex process for specifically selecting children for inclusion in the study. Nonetheless, the comparability of the results of home and sleep laboratory overnight PSG appears good; additional study using commercially available equipment in a more representative population would be helpful.

### **Treatment of OSAS**

#### *Tonsillectomy and/or Adenoidectomy*

There are many published papers, primarily case reports and case series, that support the efficacy of tonsillectomy with or without adenoidectomy as treatment for OSAS. Most of these studies use relief of snoring and other clinical symptoms as their endpoint.<sup>56,68,71–77</sup> Others cite improvement in growth,<sup>25–29</sup> behavior,<sup>19</sup> cardiovascular complications,<sup>31,33</sup> or enuresis<sup>78</sup> after surgery. Several papers suggest that adenotonsillectomy is effective treatment of OSAS even in children who are morbidly obese.<sup>29,79,80</sup> Many of these studies are anecdotal and methodologically uninterpretable; those that used PSG to document OSAS are summarized below and in Table 7.

Frank et al<sup>81</sup> were the first to use PSG to analyze the effect of surgery on OSAS in children. Of an initial group of 32 children referred for suspected OSAS, they reported on 7 who had PSG before and after adenotonsillectomy. These children had an average of 194 obstructive apneas per night preoperatively and 7 postoperatively ( $P < .025$ ). They provide no breakdown of individual cure rate. Zucconi et al,<sup>82</sup> using nocturnal or nap PSG, reported a 100% cure rate of OSAS in 29 children receiving adenotonsillectomy or adenoidectomy and monotonosillectomy and a 0% cure rate in 5 children receiving only adenoidectomy. Two more recent studies were methodologically superior regarding diagnosis of OSAS. Suen et al<sup>48</sup> reported on 69 children referred for evaluation of possible OSAS; 35 (51%) had a RDI >5 and were referred for adenotonsillectomy, and 30 had the procedure. Follow-up PSG was performed in 26; all showed improvement, although 4 (15%) still had an RDI >5. All children with persistently high RDIs continued to snore, although 3 children with RDIs that had normalized continued to snore. Thus, adenotonsillectomy resulted in a cure rate of 85%, and the absence of postoperative snoring was associated with no treatment failures (NPV of postoperative snoring = 100%), whereas 57% of children who still snored continued to have abnormal PSG results (PPV = 57%). The authors of that paper emphasized that a high preoperative RDI was a strong predictor of abnormal postoperative RDI and suggested 19.1 as a cutoff. However, their data shows that the PPV of preoperative RDI  $\geq 19.1$  for a postoperative RDI >5 was 43% and the NPV was 95%, neither of which are as high as the predictive values afforded by the presence of persistent snoring postoperatively. The findings of Nieminen<sup>56</sup> were similar, although their criteria for positive PSG results were slightly different (AHI >1). They reported a 95% cure rate for a group of 21 children after adenotonsillectomy or tonsillectomy; 1 of 5 children who continued to snore

TABLE 7. Surgical Treatment for OSAS

Author	Year	Methodology and Rating	PSG Criterion for Surgery	Number of Subjects	Results of Surgery	Comments
Eliaschar <sup>85</sup>	1980	Case series, level V	>40 apneic episodes per night	2	Complete resolution of obstruction in both patients after adenotonsillectomy	
Frank <sup>81</sup>	1983	Case series, level V	Mixed	7	Decreased number of obstructive apneas after adenotonsillectomy	Results not broken down by individual.
Zuccom <sup>82</sup>	1993	Case series, level V	AHI >1 (overnight PSG) or abnormal nap PSG	5	A: 0% cure	
Suen <sup>48</sup>	1995	Cohort, level III	AHI >5	29	Adenotonsillectomy: 100% cure	Monotonsillectomy in 19 of 29.
Wiet <sup>80</sup>	1997	Case series, level V	AHI >5	26	Adenotonsillectomy: 85% cure	All improved PPV of postoperative snoring = 57%; NPV of postoperative snoring = 100%.
Agren <sup>83</sup>	1998	Case series, level V	Nocturnal obstruction	48 31 adenotonsillectomy patients, 20 obese	Adenotonsillectomy: preoperative AHI, 23; postoperative AHI, 6 ( $P < .01$ )	12 of 20 with obesity had adenotonsillectomy alone; report did not distinguish these patients.
Shintani <sup>84</sup>	1998	Case series, level V	Not specified; those with AHI >10 were analyzed separately	20 with AI >5; 10 with AI >1; 17	After adenotonsillectomy: snoring resolved in 19 of 20, ODI nl in 20 of 20	5 had previous A. Postoperative AHI not reported.
Nieminem <sup>56</sup>	2000	Cohort, level III	AHI >2	134 74 with AHI >10	Improvement from adenotonsillectomy: 114 (75.4%) From A: 13 (84.5%)	Preoperative AHI did not predict treatment failure.
				21 2	Adenotonsillectomy or T: 95% cure; NS: 50% cure	Surgery improved all; PPV of postoperative snoring = 20%; NPV of postoperative snoring = 100%.

PSG indicates polysomnography; A, adenoidectomy; ODI, oxygen desaturation index; nl, normal; NS, no surgery.

had postoperative PSG results that remained abnormal (PPV = 20%), and none of the children who stopped snoring had abnormal PSG results (NPV = 100%). The authors pointed out that 73% of this group had previously had their adenoids removed, implying confirmation of the lack of efficacy of adenoidectomy alone for relief of OSAS. This paper also mentioned in passing that 2 children with abnormal results of PSG did not have surgery; in 1, the follow-up PSG results were unchanged, and in the other, the results had normalized. Although no generalizations can be made on the basis of these data, it represents the only published report of follow-up PSG in children with OSAS who were not treated.

Several other papers reported PSG results in association with adenotonsillectomy, but these reports were somewhat less clearly written. Wiet et al<sup>80</sup> reported a series of 48 patients in whom sleep studies were performed because of unclear history or physical findings, or complicated OSA. An AHI >5 was considered abnormal. Thirteen patients had no complicating medical factors, and of the 35 remaining, 20 were morbidly obese. All 13 uncomplicated patients had adenotonsillectomy. They had a significant decrease in mean AHI (from 23 to 6 [ $P < .01$ ]); it was not stated whether any had residual abnormal postoperative PSG results. Of the obese patients, 12 of 20 had adenotonsillectomy alone, and the rest had uvulopharyngopalatoplasty in addition. It was not specified how the decision to perform uvulopharyngopalatoplasty was made, and the report of results for this group was not broken down by surgical procedure. Mean AHI in the obese group decreased from 33 to 4 ( $P < .001$ ). Agren et al<sup>83</sup> reported on a group of 20 children with "unequivocal anamnestic nocturnal obstructive breathing." The preoperative AHI was >5 in 10 children, and the apnea index (AI) was >1 in 17. Five of these patients had an adenoidectomy in the past. The terminology used in that paper was confusing, and it was not entirely clear whether AI meant apnea index or apnea-hypopnea index. Postoperatively, no AHI (or AI) was reported; it was stated that 5 patients still had some partial obstruction postoperatively, but all had a normal oxygen desaturation index (which had been abnormal in 13 preoperatively). Shintani et al<sup>84</sup> described 134 children referred for snoring and clinical sleep apnea; 74 had a preoperative AHI >10, but for the rest of the group, the AHI was unspecified. Of this group, 114 had adenotonsillectomy, 13 had adenoidectomy, 4 had adenoidectomy with monotonosillectomy, and 3 had tonsillectomy alone, all presumably at the discretion of the surgeon. Using the authors' criterion for improvement of a postoperative decrease in AHI by 50%, 84.5% of children who had adenoidectomy and 75.4% of those who had adenotonsillectomy were improved postoperatively (difference between adenotonsillectomy and adenoidectomy,  $P = .732$ ). In contrast to the findings of Suen et al,<sup>48</sup> the preoperative AHI in this report did not predict the likelihood of treatment failure.

To summarize these studies, all of which were case series that were reported with variable rigor, it appears that adenotonsillectomy is curative in 75% to

100% of children, even if obese. The role of adenoidectomy alone is unclear. Postoperatively, children should be retested for OSAS if they continue to snore and possibly if the preoperative AHI was high.

#### *Postoperative Complications and the Need for Inpatient Monitoring*

A number of publications have catalogued postoperative complications of adenotonsillectomy in large series of patients,<sup>86–98</sup> but these will not be discussed further here. An additional large group of papers have described the risk of complications associated with outpatient adenotonsillectomy in the general population<sup>99–110</sup>; these case series have generally excluded children with upper airway obstruction from consideration and also will not be discussed further. However, several papers provide data pertaining to complications of surgery in children undergoing adenotonsillectomy for upper airway obstruction, all specifically addressing the risk of postoperative respiratory obstruction.<sup>111–118</sup> These are listed in Table 8. These authors define respiratory compromise in various ways but generally consider the need for supplemental oxygen as a minimum criterion. The papers report a wide range for the rate of postoperative respiratory complications (0%–27%), primarily because their populations include different proportions of children with neuromuscular, chromosomal, and craniofacial disorders. This variation makes the study groups too heterogeneous for pooling of the data, and their inclusion of complex patients makes them less valid in estimating the risk of postoperative respiratory compromise in the population being addressed by this practice guideline. Young age (younger than 3 years) and associated medical problems were found in most papers to define the highest risk groups. High preoperative RDI also seems to be a risk factor for postoperative complications.<sup>112,113</sup> Time to onset of respiratory compromise appears to be brief, although McColley et al<sup>112</sup> reported that 1 patient took 14 hours to manifest respiratory symptoms.

All in all, children with OSAS clearly seem to be at high risk of postoperative respiratory compromise, and increased vigilance in postoperative monitoring is warranted.

#### *Nasal CPAP*

Several papers report on the successful use of CPAP in childhood.<sup>119–124</sup> In children, CPAP is usually used when adenotonsillectomy is unsuccessful or contraindicated rather than as a primary treatment. Thus, most cases in the above reports describe children with complicated OSAS who are not the target group for this practice guideline. For example, of 80 children reported by Waters et al,<sup>120</sup> 70 had previous adenotonsillar surgery; the 10 who did not were younger than 6 months or had other significant medical conditions. Of 94 patients reported by Marcus et al,<sup>119</sup> only 2 of 18 patients whose OSAS was idiopathic (ie, not associated with another predisposing cause) had not had previous adenotonsillectomy; 1 of these patients had cystic fibrosis. All of the patients described by Guillemainault et al (1995)<sup>124</sup>

**TABLE 8.** Respiratory Compromise After Adenotonsillectomy in Children with OSAS

Author	Year	Methodology and Rating	Inclusion Criteria	Number	Rate of Respiratory Compromise	Comments
McGowan <sup>111</sup>	1992	Case series, level IV	Clinical upper airway obstruction	53	25%	Risk factors for complications were prematurity; adenoidal facies; preoperative respiratory distress.
McColley <sup>112</sup>	1992	Case series, level IV	Abnormal PSG	69	23%	Onset up to 14 hours postoperatively. Main risk factors were age and preoperative RDI.
Price <sup>113</sup>	1993	Case series, level IV	Clinical upper airway obstruction, nap PSG	160	19%	Associations with risk factors (age, preoperative PSG) asserted but not quantitated.
Rosen <sup>114</sup>	1994	Case series, level IV	Abnormal PSG	37	27%	Postoperative obstruction occurred within hours of surgery. All patients with complications were complex and had a higher mean RDI preoperatively.
Helfaer <sup>115</sup>	1996	Case series, level IV	Mild OSAS by PSG (excluded severe patients)	15	0%	No postoperative desaturation or obstruction in children with mild OSAS.
Gerber <sup>116</sup>	1996	Case series, level IV	Questionnaire	292	15% (38% if younger than 3 years)	Included complex patients. Respiratory compromise only developed in patients who snored preoperatively.
Rothschild <sup>118</sup>	1994	Case series, level IV	Clinical diagnosis	69	7%	Specific diagnostic criteria for OSAS not specified.
Biavati <sup>117</sup>	1997	Case series, level IV	Clinical diagnosis	355 23 with PSG	25% (36% with abnormal PSG)	Included complex patients. No patient with normal PSG had postoperative respiratory complications.

PSG indicates polysomnography.

were younger than 1 year. All of the patients described by Rains et al<sup>122</sup> and by Guillemineault et al (1986)<sup>121</sup> had underlying predisposing abnormalities. All of the subjects described by Tirosh et al<sup>123</sup> had previous adenotonsillectomy. These studies do confirm, however, that CPAP is efficacious in children.

## CONCLUSIONS

### Prevalence of Childhood OSAS

Snoring is a common occurrence in childhood, with reported prevalence between 3.2% and 12.1%. The prevalence of childhood OSAS is difficult to estimate, largely because published studies use different PSG criteria for its ascertainment. Reports range from 0.7% to 10.3%.

### Sequelae of Childhood OSAS

Childhood OSAS is associated with several important sequelae and complications for which prevalence is unclear because of a lack of population-based cohort studies.

#### Neurobehavioral Complications

Cross-sectional studies suggest a nearly threefold increase in behavior problems and neurocognitive abnormalities in children with sleep-disordered breathing. Most of these studies did not definitively differentiate children with PS from those with OSAS, so the true prevalence of behavior and learning problems in children with OSAS versus PS is not clear.

#### Growth Inhibition

No systematic studies exist, but case series suggest that growth (especially weight gain) accelerates after surgery for OSAS, even in children with preexisting obesity, so it appears that OSAS has an inhibitory

effect on growth. One study suggests that this effect is attributable to increased metabolic expenditures associated with OSAS.

#### Cardiovascular Complications

Cor pulmonale, right ventricular dysfunction, and pulmonary hypertension all have been reported in case reports and series, but their prevalence is unknown. These appear to be reversible after adenotonsillectomy. Systemic hypertension is a known complication of adult OSAS, and elevated diastolic blood pressure has been found in children with OSAS.

### Diagnosis of OSAS

#### Overnight Polysomnography

The gold standard for diagnosis of OSAS is overnight PSG performed in a sleep lab. Methodologic standards and population-based normal ranges have recently been published, so although older published studies reflect a problem of variability in methods and interpretation, this has diminished in recent years. However, current normative standards for PSG determination of OSAS have been chosen on the basis of statistical distribution of data, and it has not been established that those standards have any validity as predictors of the occurrence of complications. Nonetheless, at the very least, it appears that the severity of PSG abnormality is an important predictor of complications in the immediate postoperative period after adenotonsillectomy.

#### Alternatives to PSG

Clinical evaluation, including the use of questionnaires such as the one published by Brouillette et al,<sup>49</sup> has unacceptably low sensitivity and specificity for predicting OSAS. The use of home audiotaping and videotaping to supplement the clinical evaluation

has been inadequately investigated. Additional studies are necessary before any statements about their validity can be made. Pulse oximetry and nap PSG appear to have high specificity and low sensitivity, meaning that positive test results are probably true, but negative test results would need to be confirmed using overnight PSG. The comparability of the results of home and sleep laboratory overnight PSG appears good, but additional study using commercially available equipment in a representative population is necessary for confirmation.

#### Treatment of OSAS

On the basis of case series that were reported with variable rigor, it appears that adenotonsillectomy is curative in 75% to 100% of children, even if the children are obese. The role of adenoidectomy alone is unclear. Postoperatively, children should be retested for OSAS if they continue to snore and possibly if the preoperative AHI was high. Children with OSAS clearly seem to be at high risk of postoperative respiratory compromise, and increased vigilance in postoperative monitoring is warranted, particularly in those with a high preoperative RDI. CPAP is effective in children, but it is usually used when adenotonsillectomy is delayed, contraindicated, or unsuccessful rather than as a primary treatment.

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# APPENDIX 1

## OSAS LITERATURE REVIEW FORM

1. Unique identifying number: \_\_\_\_\_
2. Title: \_\_\_\_\_
3. First Author: \_\_\_\_\_
4. Reviewer's name: \_\_\_\_\_
5. Supervising committee member's name: \_\_\_\_\_

6. Include article \_\_\_\_

Exclude article \_\_\_\_

- Article was irrelevant (explain): \_\_\_\_\_  
\_\_\_\_\_
- Patients < 1 year of age: \_\_\_\_
- Patients > 18 years of age: \_\_\_\_

7. Type of article:

- review article: \_\_\_\_
- case report: \_\_\_\_
- descriptive study: \_\_\_\_
- nonrandomized historical cohort series: \_\_\_\_
- nonrandomized concurrent cohort series: \_\_\_\_
- randomized clinical trials: \_\_\_\_

8. Category

- Epidemiology (includes prevalence, risk factors): \_\_\_\_
- Diagnostic techniques (includes history, examination, special studies, polysomnography): \_\_\_\_
- Treatment (includes complications, risk factors for postoperative complications, treatment risks and benefits): \_\_\_\_
- Complications of OSAS: \_\_\_\_
- Prognosis (short- and long-term outcomes): \_\_\_\_
- Other (explain): \_\_\_\_\_

9. Hypothesis: \_\_\_\_\_

10. Number of subjects: \_\_\_\_

11. How was the study group defined? \_\_\_\_\_  
\_\_\_\_\_

12. Exclusion criteria for subjects: \_\_\_\_\_

Not mentioned: \_\_\_\_

13. Number of controls: \_\_\_\_

14. How was the control group defined? \_\_\_\_\_  
\_\_\_\_\_

15. Exclusion criteria for controls: \_\_\_\_\_

Not mentioned: \_\_\_\_

16. Describe randomization methods: \_\_\_\_\_

Not mentioned: \_\_\_\_

17. Was the study: Single-blinded: \_\_\_\_ Double-blinded \_\_\_\_ Non-blinded \_\_\_\_

18. Describe the intervention: \_\_\_\_\_  
\_\_\_\_\_

19. Definition of outcome measures: \_\_\_\_\_

20. Synopsis of methods: \_\_\_\_\_  
\_\_\_\_\_

21. Statistical methods: \_\_\_\_\_

22. Attrition rate (subjects lost to follow-up/crossed-over groups etc.): \_\_\_\_\_  
\_\_\_\_\_

23. Confounding variables: \_\_\_\_\_

24. Synopsis of results: \_\_\_\_\_  
\_\_\_\_\_

25. Were results statistically significant?

Yes \_\_\_\_ No \_\_\_\_

If yes, list positive results with *P* values: \_\_\_\_\_

---

26. For negative trials, were post hoc power analyses/confidence intervals provided?

Yes \_\_\_\_ No \_\_\_\_

If yes, give details: \_\_\_\_\_

27. Were sensitivity results provided? Yes \_\_\_\_ No \_\_\_\_

If yes, describe the measure and state the sensitivity: \_\_\_\_\_

28. Were specificity results provided? Yes \_\_\_\_ No \_\_\_\_

If yes, describe the measure and state the specificity: \_\_\_\_\_

29. Was a cost analysis provided: Yes \_\_\_\_ No \_\_\_\_

If yes, give details: \_\_\_\_\_

30. Synopsis of conclusions: \_\_\_\_\_

\_\_\_\_\_

Critique: \_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

## Technical Report: Diagnosis and Management of Childhood Obstructive Sleep Apnea Syndrome

Michael S. Schechter and Section on Pediatric Pulmonology, Subcommittee on Obstructive Sleep Apnea Syndrome  
*Pediatrics* 2002;109:e69

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