

# Sleep Apnea–Hypopnea Syndrome in a Pediatric Population: Differences Between Children With Tonsillar Hypertrophy and Those With Concomitant Disease

Mónica Llombart, Eusebi Chiner, Elia Gómez-Merino, Ada Andreu, Esther Pastor, Cristina Senent, Ana Camarasa, and Jaime Signes-Costa

Unidad de Trastornos de Sueño, Sección de Neumología, Hospital Universitari Sant Joan d'Alacant, San Juan de Alicante, Alicante. Spain

**OBJECTIVE:** Our aim was to compare clinical and polysomnographic variables in pediatric patients with sleep apnea–hypopnea syndrome (SAHS) secondary to tonsillar hypertrophy with those in patients with concomitant disease.

**PATIENTS AND METHODS:** We studied 42 children with SAHS (mean [SD] age, 8 [4] years; body mass index [BMI], 19.6 [5.2] kg/m<sup>2</sup>; neck circumference, 29 [4] cm; and BMI percentile, 67 [36]), 26 of whom were otherwise healthy (group A) and 16 of whom had concomitant disease (group B).

**RESULTS:** A comparison of groups A and B showed no significant differences in age (7.7 [3.9] years vs 8.4 [3.9] years; *P*=not significant [NS]); sex, BMI (17.6 [4] kg/m<sup>2</sup> vs 20.4 [6] kg/m<sup>2</sup>; *P*=NS), neck circumference (29.3 [4.7] cm vs 30.7 [3.5] cm; *P*=NS), or BMI percentile (61 [37] vs 76 [34]; *P*=NS). Tonsillar hypertrophy was more frequent in group A (*P*=.02) and craniofacial abnormalities (*P*=.008), macroglossia (*P*=.04), and dolichocephalia (*P*=.04) were more frequent in group B. No significant differences were observed in neurophysiologic variables or in the respiratory disturbance index, although group A presented higher oxygen saturation levels (97 [1.7] vs 95 [2]; *P*<.007), lower oxygen desaturation index scores (7 [7] vs 15 [10]; *P*=.007), and a lower cumulative percentage of time with oxygen saturation lower than 90% (2.2 [4] vs 16.4 [4]; *P*=.01). Twenty-three patients (88.5%) in group A underwent tonsillectomies compared to 7 (44%) patients in group B (*P*=.003). Seven patients (44%) in group B were treated with continuous positive airway pressure (CPAP) and 2 patients were treated with bilevel positive airway pressure (BiPAP), compared to 1 patient (3.8%) treated with CPAP in group A (*P*=.003). Three children in group B underwent maxillary surgery. The evolution of clinical and polygraphic variables was more favorable in group A (*P*=.04).

**CONCLUSIONS:** Children with SAHS suffer from repeated infections, delayed weight gain, hyperactivity, and neuropsychiatric manifestations. Obesity (associated with concomitant disease) and sleepiness are uncommon. Although most patients require surgery, as many as a third require treatment with CPAP or BiPAP. Furthermore, children with SAHS and concomitant disease show no specific clinical characteristics, although they tend to be

more obese, have more craniofacial abnormalities, and greater nocturnal hypoventilation.

**Key words:** Sleep apnea–hypopnea syndrome. Children. SAHS. Diagnosis. Treatment.

Síndrome de apneas-hipopneas durante el sueño en población infantil: diferencias en su expresión entre niños con hipertrofia amigdalар y con enfermedad concomitante

**OBJETIVO:** Comparar la expresión clínica y polisomnográfica del síndrome de apneas-hipopneas durante el sueño (SAHS) en niños con hipertrofia amigdalар y enfermedad concomitante.

**PACIENTES Y MÉTODOS:** Se estudió a 42 niños con SAHS –con una edad media (± desviación estándar) de 8 ± 4 años, índice de masa corporal (IMC) de 19,6 ± 5,2 kg/m<sup>2</sup>, cuello de 29 ± 4 cm y percentil de IMC de 67 ± 36–, 26 sanos (grupo A) y 16 con enfermedad concomitante (grupo B).

**RESULTADOS:** Al comparar los grupos A y B no se observaron diferencias en la edad (7,7 ± 3,9 frente a 8,4 ± 3,9 años; *p* = no significativa [NS]), el sexo, el IMC (17,6 ± 4 frente a 20,4 ± 6 kg/m<sup>2</sup>; *p* = NS), el perímetro del cuello (29,3 ± 4,7 frente a 30,7 ± 3,5 cm; *p* = NS) ni el percentil de IMC (61 ± 37 frente a 76 ± 34; *p* = NS). En el grupo A fue más frecuente la hipertrofia amigdalар (*p* = 0,02), y en B, las alteraciones del macizo facial (*p* = 0,008), macroglosia (*p* = 0,04) y dolicocefalia (*p* = 0,04). No se observaron diferencias en las variables neurofisiológicas ni en el índice de alteración respiratoria, aunque el grupo A presentó mayor saturación de oxígeno basal (97 ± 1,7 frente a 95 ± 2%; *p* < 0,007), menor índice de desaturaciones/h (7 ± 7 frente a 15 ± 10; *p* = 0,007) y menor porcentaje de tiempo de sueño con saturación de oxihemoglobina inferior al 90% (2,2 ± 4 frente a 16,4 ± 4; *p* = 0,01). Fueron tratados con amigdalectomía 23 pacientes del grupo A (88,5%) frente a 7 (44%) del B (*p* = 0,003). En el grupo B, 7 pacientes recibieron tratamiento con presión positiva continua de la vía aérea (44%) y 2 con BiPAP®, frente a uno (3,8%) en el grupo A (*p* = 0,003). Se realizó cirugía maxilar a 3 niños del grupo B. La evolución clínica y poligráfica fue más favorable en el grupo A (*p* = 0,04).

**CONCLUSIONES:** Los niños con SAHS cursan con infecciones de repetición, retraso ponderal, hiperactividad y manifestaciones neuropsíquicas, mientras que son poco frecuentes la somnolencia y la obesidad, la cual se asocia a enfermedad concomitante. Aunque la mayoría necesitará cirugía, hasta un tercio precisará tratamiento con presión positiva continua de la vía aérea/BiPAP®. Además, los niños

Correspondence: Dra. M. Llombart.  
Sección de Neumología, Hospital Universitari Sant Joan d'Alacant.  
Ctra. Alicante-Valencia, s/n. 03550 San Juan de Alicante. Alicante. España.  
E-mail: llombart\_mon@gva.es.

Manuscript received October 9, 2006. Accepted for publication April 3, 2007.

**con SAHS y enfermedad concomitante no muestran características especiales en su expresión clínica, aunque tienden a ser más obesos, con mayores alteraciones del macizo facial y mayor hipoventilación nocturna.**

**Palabras clave:** *Síndrome de apneas-hipopneas durante el sueño. Niños. SAHS infantil. Diagnóstico. Tratamiento.*

## Introduction

Sleep-disordered breathing, and sleep apnea–hypopnea syndrome (SAHS) in particular, constitutes a considerable problem in developed countries, where it affects about 4% of men and 2% of women in the adult population.<sup>1</sup> It is generally accepted that SAHS leads to cardiovascular morbidity in the form of hypertension, ischemic heart disease, and cerebrovascular disease,<sup>2,3</sup> mortality from traffic accidents,<sup>4</sup> and impaired cognitive function of multifactorial origin.<sup>5</sup> In recent years, attention has been centered on the child population and prevalence studies have shown that it may affect 2% of children.<sup>6</sup> In most cases, the main cause is adenotonsillar hypertrophy, but other causes, such as obesity and abnormal upper airway muscle tone, can also be found in this population. In fact, the prevalence of overweight in children and adolescents is 17% in some countries.<sup>7</sup> Certain syndromes, such as craniofacial abnormalities,<sup>8</sup> Down syndrome, and neuromuscular diseases, that affect the pediatric population may also predispose children to or cause SAHS.<sup>9</sup> SAHS constitutes an important cause of morbidity in children and is associated with neurocognitive and behavioral disorders.<sup>10</sup> The cardiovascular effects of childhood SAHS are now being recognized and some studies have shown an increased risk of cardiovascular disease, as in adults.<sup>11</sup> Furthermore, its clinical expression varies considerably, with several different manifestations.<sup>9</sup> SAHS in children is usually managed in pneumology sleep units, although most units have little experience with it.

The objectives of the present study were to evaluate the characteristics of pediatric SAHS in our sleep unit and to determine whether there were differences in the clinical, polysomnographic, or clinical expression of SAHS in children with tonsillar hypertrophy compared to those with concomitant disease.

## Patients and Methods

### *Study Period and Population*

We retrospectively reviewed the clinical and anthropometric characteristics of patients younger than 15 years who had been diagnosed with sleep-disordered breathing in our sleep unit over the last 10 years. All patients had been referred by various specialists due to the presence of snoring, apneas reported by their parents, and other manifestations related to the poor quality of nighttime sleep, whether or not accompanied by daytime neuropsychologic or neurophysiologic symptoms, such as fatigue or sleepiness.

We collected data related to anthropometric characteristics (height, weight, and neck circumference). The percentage of obese patients was taken to be the percentage of patients with a body mass index (BMI) in the 97th percentile or above, according to the Spanish reference tables for age and sex. The physical

examination of the oral cavity included the oropharyngeal Mallampati classification, the degree of narrowing of the palate, cranial morphology, and features of facial structure.

Parents were interviewed using a structured questionnaire that included information on the presence or absence of snoring, apneas, nighttime breathing difficulties, restless sleep, night sweats, enuresis, nasal obstruction, rhinorrhea, breathing through the mouth, difficulty hearing, recurrent infections, wheezing, heartburn, headaches, hyperactivity or attention deficit, apathy, shyness, sleepiness, poor school performance, failure to thrive, and other symptoms, such as poor appetite or overeating.

### *Diagnostic Protocol*

Diagnosis of SAHS was established by polysomnography, respiratory polygraphy, or pulse oximetry. For polysomnography we used a conventional polysomnograph (Sleeplab, Jaeger, Würzburg, Germany) that monitors neurophysiological variables by means of an electroencephalogram (C3–A2), electro-oculogram, and chin electromyogram, as well as respiratory variables such as nasal airflow using a nasal cannula, respiratory effort using thoracoabdominal bands, and oxygen saturation. We also monitored the electrocardiogram, body position, snoring, and tibial electromyogram.

Apnea was defined as a reduction in the amplitude of the oronasal airflow signal of more than 90%, whether or not associated with arousal or desaturation. Hypopnea was defined as a discernible decrease (>30% and <90%) in the amplitude of the oronasal airflow signal accompanied by arousal and/or a desaturation of 3%. Respiratory effort-related arousal was defined as a period of airflow limitation greater than 10 seconds and less than 2 minutes ending in an arousal.<sup>12</sup> The total number of apneas, hypopneas, and respiratory effort-related arousals were divided by total sleep time to obtain the respiratory alteration index, equivalent to the respiratory disturbance index proposed by Guilleminault et al.<sup>13</sup> Sleep stages were classified according to international guidelines.<sup>14</sup>

For respiratory polygraphy we used the previously-validated Stardust system,<sup>15</sup> (Respironics, Murrsville, California, USA), which allowed us to monitor airflow using a nasal cannula, respiratory movements using a thoracic band, oxygen saturation, heart rate, and body position. The definitions of apnea and hypopnea were the same as those used for polysomnography. However, since the respiratory polygraphy device does not allow the recording of neurological variables, a desaturation of at least 3% was required to meet the criteria for hypopnea. Finally the apnea–hypopnea index (AHI) was obtained, expressed as the total number of events divided by recording time. All signals were processed by computer and reviewed manually by a trained observer. For pulse oximetry, the Nonin 5000 oximeter (Nonin Medical Inc, Minneapolis, Minnesota, USA) was used at a sampling frequency of 5 seconds; desaturation was defined as a decrease of at least 3% with respect to the mean saturation of the previous minute. The signal was recorded on a computer system for the manual revision of the raw trace at 1-minute intervals.<sup>16</sup> In this way, the desaturation index, defined as the total number of desaturations divided by recording time, was obtained. All studies were carried out in a single bed used specifically for this purpose, with continuous monitoring by nurses.

The cutoff point for a diagnosis of SAHS was at least 3 events per hour, in accordance with the recent Spanish consensus statement on SAHS,<sup>12</sup> estimated by the respiratory alteration index, the AHI, or the desaturation index. A score of between 3 and 5 events per hour was considered to be diagnostic of mild SAHS; a score of between 5 and 20 events per hour was considered to be moderate SAHS; and a score higher than 10 events per hour was considered to be severe SAHS. A diagnosis

of hypoventilation was made when saturation was less than 90% for at least 10% of recording time and awake PaCO<sub>2</sub> was more than 45 mm Hg.

### Patient Groups

After evaluating the overall characteristics of the patients studied we carried out a comparative study of the anthropometric, clinical, and outcome characteristics, as well as of the overnight polygraphy study. For this purpose, patients were divided into 2 groups: patients diagnosed with SAHS secondary to adenotonsillar hypertrophy (group A) and patients with SAHS and concomitant disease (group B). Group B consisted of patients previously diagnosed with diseases that could predispose them to sleep-disordered breathing (abnormal facial morphology, Down syndrome, etc), but who had also been evaluated for snoring or symptoms suggestive of SAHS.

### Therapeutic Protocol

After diagnosis, patients were referred to the otorhinolaryngology department if the therapeutic approach considered was adenotonsillectomy or for maxillofacial surgery if indicated. Children who required continuous positive airway pressure (CPAP) or bilevel positive airway pressure (BiPAP) were offered the opportunity to adjust to treatment by using a nasal mask during the day for a week before beginning treatment. Once they had adjusted to the mask, a titration study was carried out in the hospital by means of polysomnography. For the follow-up, parents were interviewed again using the same questionnaire as at the beginning of the study. Children without concomitant disease underwent a control sleep study between 6 and 12 months after surgery. Children on ventilation were followed regularly in our outpatient clinics, initially on a monthly and subsequently on a quarterly basis. Polysomnography was repeated at 3 months and thereafter according to the patient's growth and clinical condition.

For the evaluation of response to treatment, symptoms and polygraphic variables (obtained by polysomnography or by in-hospital respiratory polygraphy) were assessed together. Thus, a patient was considered to have been cured when symptoms and the respiratory alteration index or the AHI score were less than 3 events per hour; to have improved when 50% of symptoms persisted and/or when the respiratory alteration index or the AHI score had fallen more than 50% from the previous reading; and not to have been cured when symptoms persisted and/or there were no changes in sleep study variables.

### Statistical Analysis

Quantitative variables were expressed as means (SD), and the frequency distribution, expressed as percentages, was obtained for qualitative variables. For the between-group comparison of qualitative variables we used the Fischer exact test and for quantitative variables we used the Student *t* test for unpaired samples or the Mann-Whitney test when samples did not follow a normal distribution. Statistical significance was set at a value of *P* less than .05. Data were analyzed with the SPSS software package, version 12 (SPSS, Inc, Chicago, Illinois, USA).

### Results

Over a period of 10 years we studied 42 patients (24 boys and 18 girls) referred to our department for suspected sleep-disordered breathing from the pediatrics department (38%), the otorhinolaryngology department (31%), the pneumology department (26%), the neurology department

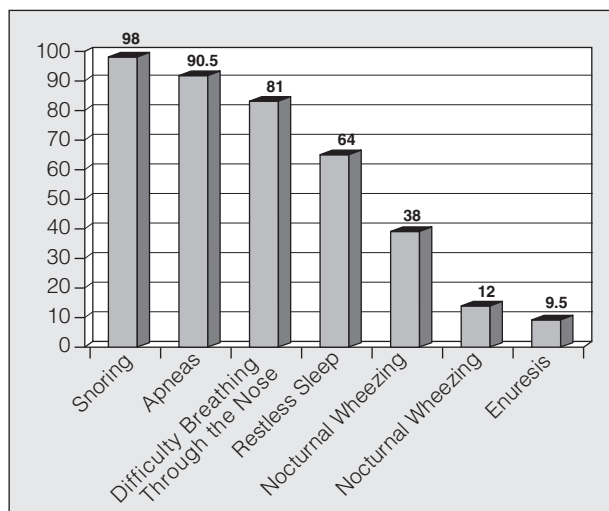


Figure 1. Nighttime symptoms of sleep apnea-hypopnea syndrome in children, in descending order of frequency

(2.5%), and the maxillofacial department (2.5%). Mean (SD) age was 8 (4) years; BMI, 19.6 (5.2) kg/m<sup>2</sup>; neck circumference, 29 (4) cm; BMI percentile; 67 (36); and weight percentile, 67 (37) (obese patients, 17%). Twenty-six of the children were healthy and 16 presented a concomitant disease: Down syndrome (n=3), Prader-Willi syndrome (n=3), cystic fibrosis (n=2), asthma (n=3), ciliary dyskinesia (n=1), morbid obesity (n=1), Pierre Robin syndrome (n=1), Rett syndrome (n=1), and neuromuscular disease (n=1).

Clinical manifestations in the patient population in descending order of frequency were snoring (98%), apneas (95%), difficulty breathing at night (81%), restless sleep (64%), recurrent upper airway infections (62%), nasal obstruction (43%), breathing through the mouth (45%), recurrent middle ear infections (41%), night sweats (38%), hyperactivity or attention deficit (31%), sleepiness (21%), headache (21%), failure to thrive (21%), poor appetite (19%), poor school performance (19%), rhinorrhea (17%), shyness (17%), difficulty hearing (17%), nocturnal wheezing (12%), enuresis (9.5%), difficulty swallowing (7%), heartburn (2.4%), and nausea and vomiting (2.4%). Figure 1 shows nighttime symptoms; Figure 2, daytime symptoms; and Figure 3, neuropsychologic symptoms. Predisposing factors were 1 or more of the following: tonsillar hypertrophy (76%), adenoid hypertrophy (48%), overweight (36%), craniofacial abnormalities (31%), high-arched palate (21.4%), macroglossia (7%), and dolichocephalia (7%). Six of the patients (14.3%) had previously undergone tonsillectomy. Seventeen percent of patients had a BMI in the 97th percentile or above for their age and sex.

For the diagnosis, 26 polysomnographies (62%), 11 in-hospital respiratory polygraphy studies (26%), and 5 pulse oximetry studies were performed. SAHS was classified as mild in 1 patient (4%), moderate in 7 patients (23%), and severe in 22 patients (73%). The final diagnosis was SAHS secondary to tonsillar or adenoid hypertrophy alone in 26 patients (62%), and SAHS caused by a combination of factors in 16 patients (38%), 3 (7%) of whom showed hypoventilation.



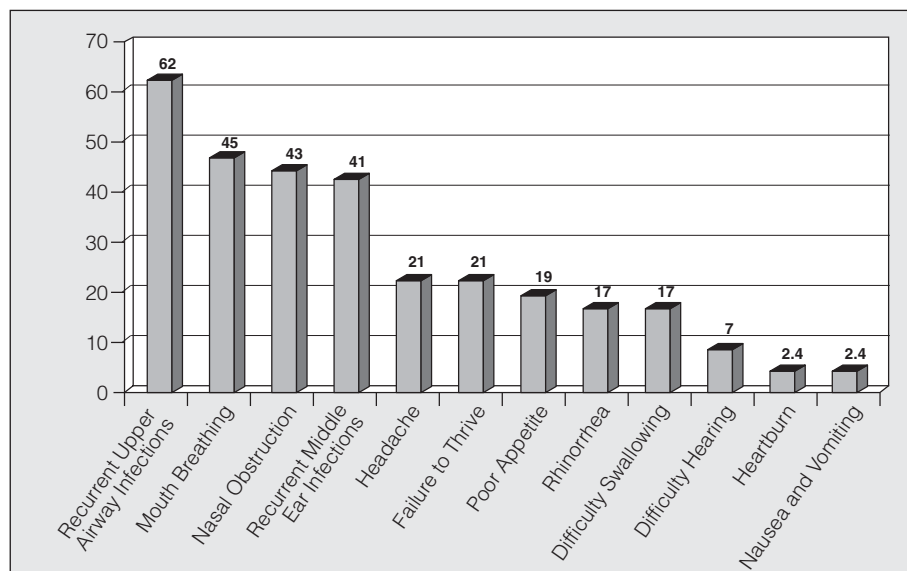


Figure 2. Daytime symptoms of sleep apnea-hypopnea syndrome in children, in descending order of frequency

TABLE 1  
Comparison of Predisposing Factors for Sleep Apnea-Hypopnea Syndrome Between Patients With Adenoid or Tonsillar Hypertrophy Alone (Group A) and Those With Concomitant Disease (Group B)\*

	Group A, %	Group B, %	P
Tonsillar hypertrophy	88	56	.02
Adenoid hypertrophy	54	37	NS
Previous tonsillectomy	12	19	NS
Craniofacial abnormalities	15	56	.008
Macroglossia	0	19	.04
Dolichocephalia	0	19	.004
Obesity	14	28	NS
High-arched palate	15	31	NS
Micrognathism	8	25	NS
Prognathism	3	12	NS
Retrognathism	18	37	NS
Dental malocclusion	4	19	NS

\*NS indicates not significant.

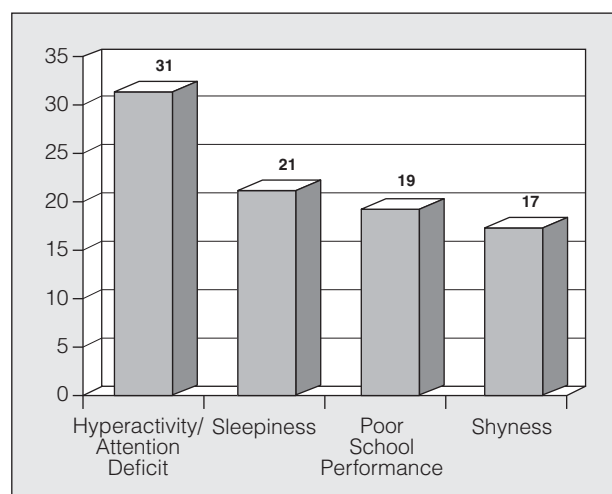


Figure 3. Neuropsychological symptoms of sleep apnea-hypopnea syndrome in children, in descending order of frequency

The therapeutic strategy was tonsillectomy with or without adenoidectomy or maxillary surgery in 71.4% of patients, CPAP in 19%, and BiPAP in the 2 patients (5%) with Prader-Willi syndrome who met criteria for associated hypoventilation. Of the 9 children treated with CPAP or BiPAP, 1 failed to adjust to treatment, 2 showed poor adherence, and 6 adhered to treatment for more than 6 hours a day. This represents an overall adherence of 60%.

Following treatment, 54% of patients were considered to have been cured, 40% were considered to have improved, and 3% were considered to have had no improvement. One patient with Prader-Willi syndrome died suddenly at night when nighttime BiPAP ventilation was momentarily abandoned. The child who did not accept treatment was lost to follow-up.

Comparison of anthropometric characteristics of group A (children with tonsillar hypertrophy only) and group B (children with concomitant disease) showed no significant differences in age (7.7 [3.9] vs 8.4 [3.9] years;  $P$ =not

significant [NS]), sex, BMI (17.6 [4] vs 20.4 [6] kg/m<sup>2</sup>;  $P$ =NS), neck circumference (29.3 [4.7] vs 30.7 [3.5] cm;  $P$ =NS), BMI percentile (61 [37] vs 76 [34];  $P$ =NS), or weight percentile (61 [42] vs 71 [33];  $P$ =NS). Although the percentage of obese patients was lower in the group with tonsillar hypertrophy (14% compared to 28% in the group with concomitant disease), this difference was not significant.

There were no differences between groups with regard to either daytime or nighttime symptoms. Comparison of predisposing factors showed that in group A tonsillar hypertrophy was more frequent, while craniofacial abnormalities and dolichocephalia were more common in group B. Table 1 shows the comparison of predisposing factors between the 2 groups. PGS showed no difference in the neurophysiological variables or in respiratory alteration index scores, although group A had greater baseline oxygen saturation, a lower desaturation index, and a lower percentage of sleep time with SpO<sub>2</sub> less than 90% (Table 2).

Twenty-three patients (88.5%) in group A underwent tonsillectomy compared to 7 (44%) in group B ( $P=.003$ ). Seven patients (44%) in group B received CPAP compared to 1 (3.8%) in group A ( $P=.003$ ). The 2 patients treated with BiPAP were also in group B. Two patients in this group underwent lower jaw surgery and 1 underwent orthodontic surgery.

The evolution of SAHS (evaluated by clinical response and nighttime polysomnography) with 1 or more combined therapies was more favorable in group A ( $P=.04$ ), although no children were considered to have been cured in either group when clinical and polysomnography criteria were applied.

### Discussion

SAHS is a well-known clinical entity in adults, but it has been studied less in the pediatric population. Childhood SAHS, which affects between 1% and 3% of children,<sup>6</sup> differs from adult SAHS both in its etiology and in its clinical manifestations, diagnosis, and treatment. Its main predisposing factor is adenotonsillar hypertrophy, although this risk factor may be favored by the increase in the prevalence of obesity in mid childhood and adolescence.<sup>7</sup> Other risk factors are craniofacial abnormalities and the anatomical narrowing of the upper airway, neuromuscular diseases, decreased upper airway muscle tone, and prematurity.<sup>17,18</sup>

Untreated SAHS is associated with neurocognitive and cardiovascular disorders. In a recent meta-analysis whose objective was to determine whether there was a relation between SAHS and cardiovascular disease in children, it was shown that the risk is increasing. However, there is still not sufficient evidence of its association with detectable abnormalities.<sup>11</sup>

Unlike the profile observed in adults, the profile of the obese child with a tendency to sleepiness and snoring corresponds to only a small percentage of children with SAHS. Most show failure to thrive and are hyperactive during the day.<sup>18</sup> In our series sleepiness was observed in only 21% of patients, while failure to thrive and behavioral disorders were more common. Nevertheless, it was recently reported that up to 43% of patients may show sleepiness if specific questionnaires are used and if the neurophysiological variables of polysomnography are analyzed.<sup>19</sup>

The natural history and long-term prognosis of pediatric SAHS are unknown, although complications may range from simple snoring to behavioral disorders, stunted growth, cor pulmonale, or even sudden death,<sup>20</sup> as occurred in 1 of our patients. Utilization of health care services among children with SAHS is high 1 year before diagnosis and treatment, and early intervention could therefore be cost effective.<sup>21</sup>

The gold standard for diagnosing SAHS is overnight polysomnography,<sup>22,23</sup> although simple diagnostic methods<sup>24,25</sup> such as the Brouillette index<sup>26</sup> have been proposed. However, the validity of this index for predicting the probability of SAHS has been questioned by other authors.<sup>24</sup> Brouillette et al<sup>27</sup> also showed that when the medical history is suggestive of SAHS, abnormal pulse

TABLE 2  
Comparison of Polysomnographic Variables Between Patients With Adenoid or Tonsillar Hypertrophy Alone (Group A) and Those With Concomitant Disease (Group B)\*

	Group A, %	Group B, %	P
Time in bed, min	478 (92)	466 (67)	NS
Sleep efficiency, %	88 (6)	83 (11)	NS
Latency to stage 1	20 (50)	40 (68)	NS
Latency to stage 2	11 (12)	9 (7)	NS
Latency to stage 3	23 (33)	23 (30)	NS
Latency to stage 4	230 (118)	202 (247)	NS
REM latency	79 (95)	44 (63)	NS
Stage 1, %	16 (17)	15 (15)	NS
Stage 2, %	31 (8)	37 (15)	NS
Stage 3, %	26 (12)	19 (12)	NS
Stage 4, %	4 (9)	7 (10)	NS
REM stage, %	23 (13)	20 (17)	NS
Arousal index	18 (11)	23 (16)	NS
RAI	18 (11)	19 (9)	NS
DI	7 (7)	15 (10)	.007
Baseline SpO <sub>2</sub> , %	97 (2)	95 (2)	.007
Minimum SpO <sub>2</sub> , %	81 (11)	79 (13)	NS
CT90%	2.2 (4)	10.3 (15.7)	.02

\*RAI indicates respiratory alteration index; NS, not significant; DI, desaturation index; SpO<sub>2</sub>, oxygen saturation as measured by pulse oximetry, CT90%, cumulative percentage of time with SpO<sub>2</sub> less than 90%. Data are shown as means (SD).

oximetry findings have a positive predictive value of 97%. However, normal pulse oximetry does not rule out mild or moderate SAHS, as only 44% of patients with SAHS diagnosed by polysomnography have abnormal pulse oximetry results.<sup>27</sup> Caution is also needed when an abbreviated test gives a positive result, as the rate of false positives is high.<sup>28</sup> Respiratory polygraphy, used in 26% of our patients, has not been fully validated in children, although some promising studies have been published.<sup>29</sup> When considered together with clinical criteria and home videotape recordings, it can constitute a good initial diagnostic approach in children with clinical suspicion of SAHS.<sup>30</sup>

In a cohort of children between 6 and 11 years it was observed that snoring, daytime sleepiness, and learning difficulties were in themselves very specific but not very sensitive for the diagnosis of sleep-disordered breathing. However, when these 3 factors were considered together, the specificity and positive predictive value were high enough to indicate that many children do not require polysomnography for diagnosis.<sup>31</sup> Our intention was to perform polysomnography, and we did so in 62% of patients; only in a few patients (12%) did we perform pulse oximetry, which was replaced in the most urgent cases by in-hospital respiratory polygraphy.

The patients with SAHS and concomitant disease merit special commentary. This group included children with asthma and cystic fibrosis, among other diseases. It has been shown that snoring and observed apneas are more prevalent in patients with asthma symptoms, and an association with sleep-disordered breathing has been suggested.<sup>32</sup> An association has also been found between obesity and wheezing, and some authors have suggested that this association must be in part mediated by factors

associated with sleep-disordered breathing, such as obesity.<sup>33,34</sup> Children with Down syndrome have a greater risk of presenting SAHS due to the conjunction of such factors as micrognathism, low muscle tone, macroglossia, and midfacial hypoplasia, together with obesity and adenotonsillar hypertrophy.<sup>34</sup> A study in which 108 children with Down syndrome underwent respiratory polygraphy showed that the prevalence of sleep-disordered breathing was 55%, and that it was higher in boys than in girls.<sup>35</sup> Similarly, in another recent study of 56 children between the ages of 2 and 4 years, 57% were diagnosed with SAHS. This percentage was as high as 80% when an abnormal arousal index score was taken into account. For this reason, it is recommended that polysomnography be performed in such children even when their parents do not report suspicion of a sleep disorder.<sup>36</sup> In patients with Prader-Willi syndrome there is a high prevalence of SAHS (AHI>10/h in 69%); obesity is common and associated with more severe sleep-related breathing disorders. Such patients present excessive daytime sleepiness, increased impulsiveness, pseudoautistic behavioral disorders,<sup>37</sup> and a wide variety of respiratory abnormalities, such as SAHS and nocturnal hypoventilation. The abnormal response to hypoxia in such patients may lead to a high morbidity rate and even to sudden death,<sup>34,37</sup> as occurred in 1 of our patients who had temporarily abandoned treatment.

The evidence concerning the clinical consequences of SAHS is overwhelming, and the "wait and see" approach is therefore not justified.<sup>30</sup> The treatment of choice is adenotonsillectomy, which is curative in most patients when there is no associated comorbidity.<sup>38-41</sup> In a recent meta-analysis of those studies in which polysomnography was performed before and after surgical treatment and from which children with significant comorbidity including craniofacial syndromes, morbid obesity, or neuromuscular diseases were excluded, treatment was shown to have been effective in 83% of cases.<sup>41</sup> However, residual SAHS may be present in a high percentage of patients, and complete resolution should therefore be checked systematically. In our study, 3 children presented residual SAHS following adenotonsillectomy. All 3 belonged, however, to the group with concomitant disease, in which the probability of residual SAHS is greater.<sup>42</sup>

In a series of patients with clinical SAHS who underwent adenotonsillectomy, it was observed that children under 3 years had about twice the risk (10%) of presenting complications than older children.<sup>43,44</sup> These data should be taken into account in evaluating whether a patient should undergo outpatient surgery or remain in hospital. Adenotonsillectomy significantly reduces health resource utilization in children with SAHS. Untreated children consume considerable resources, and their high morbidity rate is mainly attributable to upper respiratory infections (62% in our series).<sup>45</sup> Improvements in behavior and in attention capacity in children between the ages of 4 and 11 years have been observed 3 months after adenotonsillectomy.<sup>10</sup> A correlation between weight gain and a decrease in hyperactivity 1 year after surgery,<sup>40</sup> and improvement in nocturnal enuresis<sup>46</sup> have also been observed.

CPAP is indicated especially in patients who either fail to respond or respond only partially to adenotonsillectomy and in those with predisposing factors.<sup>29,38,47</sup> In a retrospective study of 80 children treated with CPAP, many of them with congenital syndromes or malformations, treatment was deemed effective in 90% of patients and was, in general, well tolerated.<sup>48</sup> Marcus et al<sup>47</sup> found that both CPAP and BiPAP were extremely effective in pediatric SAHS but were associated with a high treatment discontinuation rate (one third of patients). Even in those children who adhered to therapy, the number of hours of use was suboptimal (5.3 hours, on average). Adherence to treatment in our series was 60%, which was in line with the literature.

In cases of SAHS secondary to mandibular hypoplasia, long-term improvement in SAHS was observed after mandibular distraction surgery,<sup>8</sup> as occurred in 2 of our patients. In 1 patient with Pierre Robin syndrome, however, initial treatment was nasal CPAP, with optimal adaptation and adherence, until the patient had grown sufficiently to undergo surgery 4 years later.

The continued presence of events following surgery is well known and, as many patients present complex syndromes or obesity, we feel that it is necessary to use some diagnostic technique, even if it is an abbreviated one, to determine the need for complementary treatment.<sup>49</sup> While polysomnography was used in the majority of cases in our study, respiratory polygraphy will probably be sufficient in the future.

The present study has some potential limitations. Due to the use of different diagnostic methods, the results may not be homogeneous. In the majority of patients, polysomnography was used and the differences that may exist between the various methods were for the most part counteracted by the use of a common respiratory event index for the 3 diagnostic methods used. While our study was a retrospective one, the same protocol of data collection was used over the years and applied by the 2 principal investigators, who reviewed the overnight polygraphy studies manually.

From our study we conclude that children with SAHS suffer from repeated infections, failure to thrive, hyperactivity, and neuropsychiatric manifestations, while obesity (which is associated with concomitant disease) and sleepiness are very uncommon. While most patients require surgery for resolution, as many as a third require treatment with CPAP or BiPAP. Children with SAHS and concomitant disease show no specific clinical characteristics, although they do tend to be more obese, have more pronounced craniofacial abnormalities, and greater nocturnal hypoventilation. They often require a combination of therapies, such as CPAP or BiPAP together with surgery.

## REFERENCES

1. Young T, Palta M, Dempsey J, Skatrud J, Weber S, Badr S. The occurrence of sleep-disordered breathing among middle-aged adults. *N Engl J Med.* 1993;328:1230-5.
2. Shamsuzzaman AS, Gersh BJ, Somers VK. Obstructive sleep apnea: implications for cardiac and vascular disease. *JAMA.* 2003;290:1906-14.

3. Terán Santos J, Alonso Álvarez ML, Cordero Guevara J, Ayuela Azcárate JM, Montserrat Canal JM. Síndrome de apnea del sueño y el corazón. *Rev Esp Cardiol*. 2006;59:718-24.
4. Terán-Santos J, Jiménez-Gómez A, Cordero-Guevara J, and the Cooperative Group Burgos-Santander. The association between sleep apnea and the risk of traffic accidents. *N Engl J Med*. 1999; 340:847-51.
5. Suratt PM, Peruggia M, D'Andrea L, Diamond R, Barth JT, Nikova M, et al. Cognitive function and behavior of children with adenotonsillar hypertrophy suspected of having obstructive sleep-disordered breathing. *Pediatrics*. 2006;118:e771-e81.
6. Redline S, Tishler PV, Schluchter M, Aylor J, Clark K, Graham G. Risk factors for sleep-disordered breathing in children. Associations with obesity, race, and respiratory problems. *Am J Respir Crit Care Med*. 1999;159:1527-32.
7. Ogden CL, Carroll MD, Curtin LR, McDowell MA, Tabak CJ, Flegal KM. Prevalence of overweight and obesity in the United States, 1999-2004. *JAMA*. 2006;295:1549-55.
8. Lin SY, Halbower AC, Tunkel DE, Vanderkolk C. Relief of upper airway obstruction with mandibular distraction surgery: long-term quantitative results in young children. *Arch Otolaryngol Head Neck Surg*. 2006;132:437-41.
9. Sheldon SH, Ferber R, Kryger MH, editors. Principles and practice of pediatric sleep medicine. Philadelphia: Elsevier Saunders Inc.; 2005.
10. Galland BC, Dawes PJ, Tripp EG, Taylor BJ. Changes in behavior and attentional capacity after adenotonsillectomy. *Pediatr Res*. 2006;59:711-6.
11. Ng DK, Chan C, Chow AS, Chow P, Kwok K. Childhood sleep-disordered breathing and its implications for cardiac and vascular diseases. *J Paediatr Child Health*. 2005;41:640-6.
12. Grupo Español de Sueño (GES). Consenso Nacional sobre el síndrome de apneas-hipopneas durante el sueño. *Arch Bronconeumol*. 2005;41 Extraordinario 4;1-110.
13. Guilleminault C, Li K, Khramitsov A, Palombini I, Pelayo R. Breathing patterns in pre-pubertal children with sleep disordered breathing. *Arch Pediatr Adolesc Med*. 2004;158:153-61.
14. Rechtschaffen A, Kales A. A manual of standardised terminology, techniques and scoring system for sleep stages of human subjects. Washington DC: National Institutes of Health; 1968 [publication No. 204].
15. Chiner E, Signes-Costa J, Arriero JM, Gómez-Merino E, Andreu AL, Pastor E, et al. Validation of a portable sleep monitoring device (Stardust®) for home studies. *Am J Respir Crit Care Med*. 2003; 167:A403.
16. Chiner E, Signes-Costa J, Arriero JM, Marco J, Fuentes I, Sergado A. Nocturnal oximetry for the diagnosis of the sleep apnoea hypopnoea syndrome: a method to reduce the number of polysomnographies? *Thorax*. 1999;54:968-71.
17. Nixon GM, Brouillette RT. Sleep. 8: paediatric obstructive sleep apnoea. *Thorax*. 2005;60:511-6.
18. Brouillette RT, Fernbach SK, Hunt CE. Obstructive sleep apnea in infants and children. *J Pediatr*. 1982;100:31-40.
19. Chervin RD, Weatherly RA, Ruzicka DL, Burns JW, Giordani BJ, Dillon JE, et al. Subjective sleepiness and polysomnographic correlates in children scheduled for adenotonsillectomy vs other surgical care. *Sleep*. 2006;29:495-503.
20. Singer LP, Saenger P. Complications of pediatric obstructive sleep apnea. *Otolaryngol Clin North Am*. 1990;23:665-76.
21. Reuveni H, Simon T, Tal A, Elhayany A, Tarasiuk A. Health care services utilization in children with obstructive sleep apnea syndrome. *Pediatrics*. 2002;110:68-72.
22. American Thoracic Society. Standards and indications for cardiopulmonary sleep studies in children. *Am J Respir Crit Care Med*. 1996;153:866-78.
23. Section on Pediatric Pulmonology, Subcommittee on Obstructive Sleep Apnea Syndrome. American Academy of Pediatrics. Clinical practice guideline: diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics*. 2002;109: 704-12.
24. Villa Asensi J, De Miguel Díez J, Romero Andújar F, Campelo Moreno O, Sequeiros González A, Muñoz Codoceo R. Utilidad del índice de Brouillette en el diagnóstico del síndrome de apnea obstructiva del sueño en niños. *An Esp Pediatr*. 2000;53:547-52.
25. Kotagal S. Childhood obstructive sleep apnoea. *BMJ*. 2005;330: 978-9.
26. Brouillette R, Hanson D, David R, Klemka L, Szatkowski A, Fernbach S, et al. A diagnostic approach to suspected obstructive sleep apnea in children. *J Pediatr*. 1984;105:10-4.
27. Brouillette RT, Morielli A, Leimanis A, Waters KA, Luciano R, Ducharme FM. Nocturnal pulse oximetry as an abbreviated testing modality for pediatric obstructive sleep apnea. *Pediatrics*. 2000;105:405-12.
28. Kirk VG, Bohn SG, Flemons WW, Remmers JE. Comparison of home oximetry monitoring with laboratory polysomnography in children. *Chest*. 2003;124:1702-8.
29. Villa Asensi JR, De Miguel Díez J. Síndrome de apnea obstructiva del sueño en la infancia. *An Esp Pediatr*. 2001;54:58-64.
30. Durán Cantolla J, Rubio Aramendi R. Síndrome de apnea-hipopnea del sueño en niños. *An Esp Pediatr*. 2001;54:4-6.
31. Goodwin JL, Kaemingk KL, Mulvaney SA, Morgan WJ, Quan SF. Clinical screening of school children for polysomnography to detect sleep-disordered breathing – the Tucson Children's Assessment of Sleep Apnea Study (TuCASA). *J Clin Sleep Med*. 2005;1:247-54.
32. Ekici A, Ekici M, Kurtipek E, Keles H, Kara T, Tunckol M, et al. Association of asthma-related symptoms with snoring and apnea and effect on health-related quality of life. *Chest*. 2005;128:3358-63.
33. Sulit LG, Storfer-Isser A, Rosen CL, Kirchner HL, Redline S. Associations of obesity, sleep-disordered breathing, and wheezing in children. *Am J Respir Crit Care Med*. 2005;171:659-64.
34. Deane S, Thomson A. Obesity and the pulmonologist. *Arch Dis Child*. 2006;9:188-91.
35. de Miguel-Díez J, Villa-Asensi JR, Álvarez-Sala JL. Prevalence of sleep-disordered breathing in children with Down syndrome: polygraphic findings in 108 children. *Sleep*. 2003;26:1006-9.
36. Shott SR, Amin R, Chini B, Heubi C, Hotze S, Akers R. Obstructive sleep apnea: should all children with Down syndrome be tested? *Arch Otolaryngol Head Neck Surg*. 2006;132:432-6.
37. O'Donoghue FJ, Camfferman D, Kennedy JD, Martin AJ, Couper T, Lack LD, et al. Sleep-disordered breathing in Prader-Willi syndrome and its association with neurobehavioral abnormalities. *J Pediatr*. 2005;147:823-9.
38. McNamara F, Sullivan CE. Treatment of obstructive sleep apnea syndrome in children. *Sleep*. 2000;23 Suppl 4:142-6.
39. Sánchez Armengol A, Capote Gil F, Cano Gómez S, Carmona Bernal C, García Díez E, Castillo Gómez J. Tratamiento quirúrgico de la hipertrofia adenoamigdalares en niños con trastornos respiratorios del sueño: cambios en los patrones polisomnográficos. *Arch Bronconeumol*. 1997;33:124-8.
40. Roemlich JN, Barkley JE, D'Andrea L, Nikova M, Rogol AD, Carskadon MA, et al. Increases in overweight after adenotonsillectomy in overweight children with obstructive sleep-disordered breathing are associated with decreases in motor activity and hyperactivity. *Pediatrics*. 2006;117:200-8.
41. Brietzke SE, Gallagher D. The effectiveness of tonsillectomy and adenoidectomy in the treatment of pediatric obstructive sleep apnea/hypopnea syndrome: a meta-analysis. *Otolaryngol Head Neck Surg*. 2006;134:979-84.
42. Wiet GJ, Bower C, Seibert R, Griebel M. Surgical correction of obstructive sleep apnea in the complicated pediatric patient documented by polysomnography. *Int J Pediatr Otorhinolaryngol*. 1997;41:133-43.
43. Statham MM, Elluru RG, Buncher R, Kalra M. Adenotonsillectomy for obstructive sleep apnea syndrome in young children: prevalence of pulmonary complications. *Arch Otolaryngol Head Neck Surg*. 2006;132:476-80.
44. Nixon GM, Kermack AS, Davis M, Manoukian J, Brown KA, Brouillette RT. Planning adenotonsillectomy in children with obstructive sleep apnea: the role of overnight oximetry. *Pediatrics*. 2004;113:19-25.
45. Tarasiuk A, Simon T, Tal A, Reuveni H. Adenotonsillectomy in children with obstructive sleep apnea syndrome reduces health care utilization. *Pediatrics*. 2004;113:351-6.
46. Firoozi F, Batniji R, Aslan AR, Longhurst PA, Kogan BA. Resolution of diurnal incontinence and nocturnal enuresis after adenotonsillectomy in children. *J Urol*. 2006;175:1885-8.
47. Marcus CL, Rosen G, Ward SL, Halbower AC, Sterni L, Lutz J, et al. Adherence to and effectiveness of positive airway pressure therapy in children with obstructive sleep apnea. *Pediatrics*. 2006;117:442-51.
48. Waters KA, Everett FM, Bruderer JW, Sullivan CE. Obstructive sleep apnea: the use of nasal CPAP in 80 children. *Am J Respir Crit Care Med*. 1995;152:780-5.
49. Goldstein NA, Pugazhendhi V, Rao SM, Weedon J, Campbell TF, Goldman AC, et al. Clinical assessment of pediatric obstructive sleep apnea. *Pediatrics*. 2004;114:33-43.