

Reliability of Respiratory Polygraphy for the Diagnosis of Sleep Apnea-Hypopnea Syndrome in Children

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OBJECTIVE: Overnight polysomnography (PSG) is the gold standard diagnostic tool for sleep apnea-hypopnea syndrome (SAHS) in children. The aim of the present study was to evaluate the usefulness of diagnostic respiratory polygraphy in children with clinically suspected SAHS referred to our sleep-disordered breathing clinic.

PATIENTS AND METHODS: We studied 53 children referred with clinical suspicion of SAHS; 29 (54.7%) were boys and the mean (SD) age was 6.4 (2.9) years. After a medical history was taken and a physical examination performed, patients underwent respiratory polygraphy (Edentec) simultaneously with overnight PSG in the sleep laboratory. The 2 diagnostic tools were compared using statistical analysis.

RESULTS: SAHS was defined by an obstructive apnea-hypopnea index (OAH) of 3 or more in overnight PSG and a respiratory disturbance index (RDI) of 3 or more in respiratory polygraphy. The rate of diagnostic agreement was 84.9%. The difference between the mean OAH and RDI values was not significant (0.7 [5.4]; $P=0.34$). The intraclass correlation coefficient between the OAH and RDI was 89.4 (95% confidence interval, 82.4-93.7; $P<0.001$).

When receiver operating characteristic curves were calculated for the OAH cutoff points used for the diagnosis of SAHS (1, 3, and 5), the best RDI cutoff for all 3 OAH values considered was found to be 4.6. When age strata were considered, in children 6 years or older the best RDI cutoff for the 3 OAH values was 2.1. In children younger than 6 years the best RDI cutoff was 3.35 for OAH 1 and 5.85 for OAH 3 and 5.

CONCLUSIONS: Respiratory polygraphy in the sleep laboratory is a valid method for the diagnosis of SAHS in children.

Key words: Sleep-disordered breathing. Children. Sleep. Obstructive sleep apnea. Apnea. Sleep studies.

Fiabilidad de la poligrafía respiratoria para el diagnóstico del síndrome de apneas-hipopneas durante el sueño en niños

OBJETIVO: La polisomnografía (PSG) nocturna es la técnica diagnóstica de referencia del síndrome de apneas-hipopneas durante el sueño (SAHS) en niños. El objetivo del estudio ha sido evaluar la utilidad diagnóstica de la poligrafía respiratoria (PR) en niños con sospecha clínica de SAHS remitidos a la Unidad de Trastornos Respiratorios del Sueño.

PACIENTES Y MÉTODOS: Se estudió a 53 niños remitidos por sospecha clínica de SAHS (29 varones; 54,7%), con una edad media \pm desviación estándar de 6,4 \pm 2,9 años. A todos ellos se les realizaron historia clínica, exploración física, PR (Edentec®) y PSG nocturna simultáneamente en el laboratorio de sueño. Se realizó el análisis estadístico para comparar ambas técnicas diagnósticas.

RESULTADOS: Definiendo el diagnóstico de SAHS como la presencia de un índice de apneas-hipopneas obstructivas (IAHO) igual o mayor de 3 en la PSG y un índice de eventos respiratorios (IER) de 3 o superior en la PR, la coincidencia diagnóstica fue del 84,9%. La diferencia de medias entre el IAHO y el IER no fue significativa (0,7 \pm 5,4; $p = 0,34$). El coeficiente de correlación intraclass entre el IAHO y el IER fue de 89,4 (intervalo de confianza del 95%, 82,4-93,7; $p < 0,001$).

Para el diagnóstico de SAHS se consideraron los valores de IAHO iguales o mayores de 1; iguales o mayores de 3, e iguales o mayores de 5. Se calcularon las curvas de eficacia diagnóstica para cada uno de ellos y 4,6 resultó ser el mejor IER para los 3 valores de IAHO considerados. Al analizar por estratos de edad, en niños de 6 años o más el mejor IER obtenido para los 3 valores de IAHO considerados fue 2,1. En niños menores de 6 años se obtuvieron los siguientes valores de IER: 3,35 para IAHO de 1 o superior y 5,85 para IAHO de 3 o mayor y de 5 o superior.

CONCLUSIONES: La PR realizada en el laboratorio de sueño es un método válido para el diagnóstico de SAHS en niños.

Palabras clave: Trastornos respiratorios del sueño. Niños. Sueño. Apnea obstructiva del sueño. Apnea. Estudios de sueño.

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Introduction

Sleep apnea-hypopnea syndrome (SAHS) in children is a respiratory sleep disorder characterized by partial obstruction or intermittent total obstruction (obstructive apneas) of the upper airway; SAHS interferes with ventilation and disturbs normal sleep architecture.^{1,2} Symptoms include habitual snoring, difficulty sleeping, and behavioral problems. In childhood, SAHS is associated with considerable morbidity, as it can lead to delayed growth and development, pulmonary hypertension, behavioral abnormalities, and poor school performance.^{3,4}

Overnight polysomnography (PSG) is currently considered the gold standard diagnostic technique.⁵⁻⁷ However, procedures for performing and analyzing PSG are still insufficiently standardized.^{2,8} Meanwhile, although respiratory polygraphy is an accepted method of diagnosing SAHS in adults,⁹ its use in children has not been properly validated and research on the subject has been scant.¹⁰⁻¹³

The main aim of our study was to evaluate the diagnostic usefulness of respiratory polygraphy in children with clinical suspicion of SAHS. To that end, we calculated the respiratory disturbance index (RDI) obtained from respiratory polygraphy performed in the sleep laboratory and compared it to the obstructive apnea-hypopnea index (OAHI) determined by overnight PSG, performed simultaneously.

Patients and Methods

Study Population

We carried out a prospective, blinded, simultaneous validation study enrolling 55 children, 53 of whom completed the study. All had been referred by the otorhinolaryngology and pediatrics departments to our sleep-disordered breathing clinic in Burgos, Spain, with clinical suspicion of SAHS between July 2003 and September 2004. The sample consisted of children of both sexes aged between 2 and 14 years with signs and symptoms of SAHS (nighttime snoring and respiratory pauses). Patients with severe concomitant disease, whether medical or psychiatric, were excluded, as were those presenting symptoms of sleep disorders other than SAHS. Informed consent was provided in all cases and the study protocol was approved by the ethics committee of the Hospital General Yagüe in Burgos.

Methods

For all patients enrolled in the study, a clinical history was taken. Patients also underwent a general physical examination as well as a specific ear, nose, and throat examination. Age, weight, height, and sex were recorded. Tonsillar hypertrophy was graded as follows: grade I, 0% outside the tonsillar fossa; grade II, occupying 25% of the space between pillars; grade III, 50% of the space; and grade IV, 100% of the space.

Respiratory polygraphy and PSG were performed simultaneously in the sleep laboratory and monitored between 10 PM and 8 AM the following morning. For overnight PSG, we used the Deltamed Coherence 3NT version 3.0 polysomnograph (Diagniscan, SA; ACH-Werfen Company; Paris, France), which recorded 2 electroencephalogram channels (C3-A2, O2-A1), right and left electrooculogram, tibial and submental electromyogram, electrocardiogram, airflow measured with an oronasal thermistor, chest and abdominal movements using thoracic bands, body position by a position sensor, arterial oxygen

saturation by pulse oximetry (SpO₂), snoring and airflow using a nasal cannula. The PSG was analyzed manually using the conventional criteria of Rechtschaffen and Kales¹⁴ and arousals using the criteria of the American Academy of Sleep Medicine.¹⁵ Respiratory events were analyzed manually according to the criteria of the American Thoracic Society.^{5,6}

For respiratory polygraphy, we used the Edentec Monitoring System (Edentrace II, Model 3711; Edentec Corporation, Nellcor Puritan Bennett, Eden Prairie, Minnesota, USA), which recorded 6 channels: oronasal flow using a thermistor, chest movements by impedance plethysmography, body position by a position sensor, snoring by microphone, and heart rate and SpO₂. The respiratory polygraphy outputs were analyzed manually by the same investigator, who was blinded to the results obtained by PSG.

For both PSG and respiratory polygraphy, apnea was defined as the absence of oronasal airflow lasting at least 6 seconds (equivalent to 2 breathing cycles), either with chest and abdominal movements (obstructive apnea) or without (central apnea). Hypopnea was defined as a decrease of at least 50% in the amplitude of respiratory flow as measured with a thermistor lasting at least 6 seconds with respiratory movements, accompanied by a decrease in SpO₂ of at least 3% (in respiratory polygraphy) or by arousals (in PSG).^{15,16}

In PSG, the apnea-hypopnea index (AHI) was defined as the number of apneas and hypopneas per hour of sleep, including central apneas, and the obstructive apnea-hypopnea index (OAHI) as the number of apneas and hypopneas per hour of sleep, not including central apneas. In respiratory polygraphy, the RDI was defined as the total number of respiratory events (apneas and hypopneas) divided by total study time. The interruption of oronasal airflow that frequently follows body movements and artifacts due to motion were not counted in PSG or respiratory polygraphy.

Statistical Analysis

A descriptive analysis was performed using frequency distributions for qualitative variables, and means (SD) for quantitative variables. Means were compared using the *t* test for paired samples. We also calculated the intraclass correlation coefficient, which is useful for evaluating agreement between different measurements of a numerical variable, as it measures the proportion of total intraindividual variance. As for all proportions, intraclass correlation coefficient values can range from 0 to 1, with the greatest possible agreement corresponding to a value of 1. Agreement is considered good when the coefficient is between 0.71 and 0.9.

We also used the Bland and Altman¹⁷ method, a simple procedure to assess the agreement between 2 systems of measurement. It consists of plotting the differences between 2 measurements against their mean. If there is no systematic error, the points will be distributed randomly on either side of the line corresponding to 0 on the axis representing the difference in the means.

Receiver operating characteristic (ROC) curves, which give a global representation of diagnostic accuracy, were also calculated. The area under the curve (AUC) is the best indicator of the overall accuracy of a diagnostic test; maximum accuracy corresponds to an AUC of 1, and minimum accuracy to 0.5. We performed logistic regression analysis, which is used to assess the role of possibly relevant variables as potential modifiers of the probability of having a certain disease when both a gold standard test and an alternative diagnostic test exist.

For the analysis of validity, the following OAHI cutoff points were used: ≥ 1 , ≥ 3 , and ≥ 5 . Statistical significance was set at $P < .05$ and 95% confidence intervals (CI) were calculated. The statistical analysis was carried out with the SPSS statistical package, version 12.0 (Chicago, Illinois, USA).

TABLE 1
Neurophysiologic Parameters of the Children Studied (n=53)

Parameter	Mean (SD)
TST, min	419.4 (44.7)
Sleep efficiency, %	85.8 (7.9)
Stage I, % TST	4.2 (3.4)
Stage II, % TST	46.6 (6.8)
Stage III, % TST	11.9 (4.1)
Stage IV, % TST	16.5 (6.9)
REM stage, % TST	20.6 (5.4)
Arousal index	8.1 (6.3)

Abbreviations: REM, rapid eye movement; TST, total sleep time.

Results

Of the 55 children studied, 2 were excluded from analysis. In the first case, exclusion was due to a technical failure and a consequent polygraphy recording of only 4 hours. The second child left the sleep laboratory at 3 AM and was thus also excluded. Thus, 53 children completed the study (29 boys; 54.7%) ranging in age from 2 to 13 years, with a mean age of 6.4 (2.9) years. Four children (7.5%) had a body mass index (BMI) above the 95th percentile, and 13 (24%) had a BMI below the 50th percentile; the mean percentile was 61.7 (30.8) and the mean BMI was 18.2 (2.7) kg/m². All the children had a history of pharyngeal or tonsillar infections and 17% had a family history of SAHS.

Snoring and breathing through the mouth at night were the most frequent manifestations (92.5%). Other symptoms were nighttime (90.6%) and daytime (83%) nasal congestion, nighttime respiratory pauses (73.6%), restless sleep (66%), nighttime sweating (52.8%), daytime fatigue (39.6%), hyperactivity (30.2%), sleepiness (26.4%), enuresis (15.1%), aggressiveness (15.1%), difficulty falling asleep (13.2%), and attention deficit (9.4%).

The most frequent finding in the ear, nose, and throat examination was a decrease in pharyngeal space secondary to tonsillar hypertrophy, which was present

in 100% of patients. Tonsillar hypertrophy was grade I in 1.9% of patients, grade II in 22.6%, grade III in 43.4%, and grade IV in 32.1%. No association was found between tonsillar size and the presence of SAHS (defined as OAHl ≥ 3).¹⁸

Total study time was 488.5 (17.8) minutes, and total PSG sleep time was 419.4 (44.7) minutes. Mean values for the neurophysiologic parameters are shown in Table 1. The prevalence of SAHS according to the OAHl cutoff points used (≥ 1 , ≥ 3 , and ≥ 5) was 77.4%, 58.5%, and 50.9%, respectively. When the values recorded by polygraphy and PSG were compared (Table 2), the correlations obtained were significant for all the matched pairs and no significant differences were found in the number of respiratory events (8.3 [47.8]; $P > .05$). Defining an OAHl of 3 or higher in PSG and an RDI of 3 or higher in respiratory polygraphy as diagnostic of SAHS, we found the rate of diagnostic agreement to be 84.9%. The difference between mean OAHl and RDI values was not significant (0.7 [5.4]; $P = .34$). The intraclass correlation coefficient between the 2 was 89.4 (95% CI, 82.4-93.7; $P < .001$).

We used a Bland-Altman plot (Figure) for OAHl and RDI to assess agreement between PSG and respiratory polygraphy. All but one of the values fell within the 95% CI.

ROC Curves

For a diagnosis of SAHS, OAHl cutoff points of ≥ 1 , ≥ 3 , and ≥ 5 were used. When ROC curves were calculated for each of them, the best RDI cutoff in relation to all 3 OAHl cutoffs considered was found to be 4.6, with an AUC greater than 85% and a specificity as high as 91.7% (Table 3).

When the study sample was stratified by age, the best RDI cutoff in relation to all 3 OAHl values was 2.1 for children 6 years or older, with an AUC greater than 90% and a specificity of 100% (Table 3). In children younger than 6 years, the best RDI cutoff was 3.35 in relation to an OAHl ≥ 1 , and 5.85 for an OAHl ≥ 3 and ≥ 5 (Table 3).

TABLE 2
Mean (SD) Results of Respiratory Polygraphy (RP) and Polysomnography (PSG) in the Children Studied (n=53)

	RP	PSG	Difference in Means	Correlation ^a
Number of respiratory events	74 (108.8)	65.7 (76.7)	8.3 (47.8)	0.92
Central apneas	6.6 (6.6)	11.8 (12.5)	5.3 (9.1) ^b	0.71 ^b
Obstructive apneas	10.7 (28.5)	31.6 (47.1)	20.9 (29.5) ^b	0.81 ^b
Mixed apneas	3.2 (5.6)	0.6 (1.6)	2.5 (5.6) ^c	0.09
Hypopneas	53.5 (77.9)	16.9 (24.6)	36.6 (58.3) ^b	0.855 ^b
Obstructive apneas RDI/obstructive apneas AHI	7.9 (12.5)	8.1 (10.5)	0.2 (5.4)	0.90 ^b
Central apneas RDI/central apneas AHI	0.8 (0.8)	1.7 (1.9)	0.9 (1.4)	0.70 ^b
AHI in REM stage		11.9 (17.6)		
SpO ₂				
Baseline	97.7 (1.1)	97.7 (1.1)		
Mean	97.5 (1.5)	97.6 (1.5)	0.1 (1.1)	0.73 ^b
Minimum	91.7 (5.8)	88.7 (7.9)	2.8 (4.9) ^b	0.78 ^b
CT90%	1.1 (4.4)	1.4 (4.9)	0.3 (3.1)	0.79 ^b

Abbreviations: AHI, apnea-hypopnea index (per hour of sleep in PSG); CT90%, cumulative percentage of sleep time with SpO₂ less than 90%; RDI, respiratory disturbance index (number of apneas and hypopneas divided by total study time in respiratory polygraphy); PSG, polysomnography; SpO₂, oxygen saturation.

^aPearson's correlation coefficient. ^b $P < .001$. ^c $P = .002$.

Logistic Regression Analysis

A logistic regression model was constructed for the 3 OAHl cutoffs used (≥ 1 , ≥ 3 , and ≥ 5) with the following covariables: extreme BMI percentile (below the 5th percentile or above the 95th percentile), snoring, respiratory pauses, sleepiness, hyperactivity, and dichotomized RDI (< 4.6 or ≥ 4.6). These covariables were chosen from among all the variables recorded in the contingency tables previously constructed for each of the variables in relation to OAHl cutoffs of ≥ 1 , ≥ 3 , and ≥ 5 . The overall model was significant in all cases, but the only variable that was significant in all cases and with all models was the dichotomized RDI. The regression models did not improve the sensitivity and specificity obtained based on the ROC curves.

Discussion

SAHS is a common disease in children, with an estimated prevalence of about 5%.¹⁹ Although children with sleep-disordered breathing are relatively heavy consumers of health services, the disease is still underdiagnosed.²⁰ The condition is also associated with considerable morbidity,^{3,21} and early diagnosis therefore has important implications for prevention. Finding efficient diagnostic alternatives to PSG can improve accessibility to diagnosis and treatment for children with SAHS. According to existing data, the cutoff point most often considered normal is an OAHl of 1. However, the clinically significant cutoff is yet to be determined and there is no consensus concerning the diagnosis of childhood SAHS.^{5,16} Normal PSG values in children have recently been published.^{7,22,23}

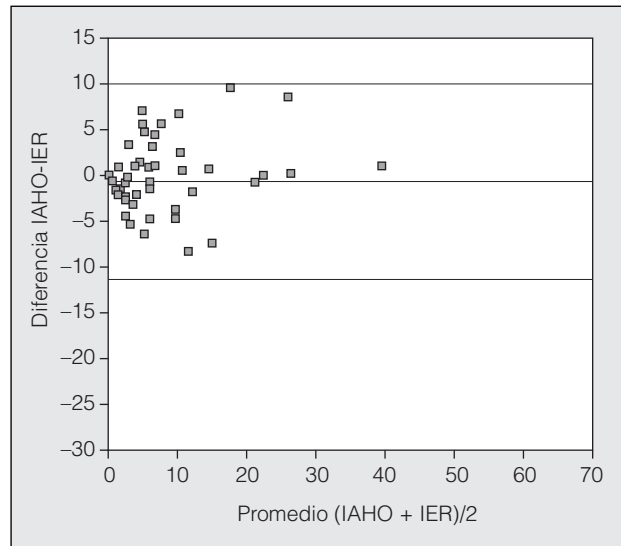


Figure. Bland-Altman plot showing agreement between respiratory polygraphy and polysomnography. As can be observed, the values fall within the limits of agreement. Abbreviations: OAHl, obstructive apnea-hypopnea index (in polysomnography); RDI, respiratory disturbance index (in respiratory polygraphy).

Several studies have attempted to find diagnostic alternatives to overnight PSG, such as the use of questionnaires based on the range of clinical manifestations, audiovisual recordings, nighttime pulse oximetry, or PSG during a daytime nap.²⁴⁻²⁷ Nevertheless, in view of the fact that the results of the studies performed may lead to surgical intervention as treatment of choice (adenotonsillectomy), and of the wide variability of the results obtained with low

TABLE 3
Diagnostically Useful RDI Values for Different Obstructive Apnea-Hypopnea Index Cutoff Points

	AUC	Sensitivity	Specificity	PP	PPP	NPP
Children aged 2-14 y (n=53)						
OAHl ≥ 1 RDI=4.6	86.2 (5.4)	63.4	91.7 77.4	96.3	57.6	
OAHl ≥ 3 RDI=4.6	85.2 (5.2)	74.2	81.8	58.5	85.2	30.7
OAHl ≥ 5 RDI=4.6	87.6 (4.9)	81.5	80.8	50.9	81.5	19.1
Children aged ≥ 6 y (n=20)						
OAHl ≥ 1 RDI=2.1	97.3 (3.3)	93.3	100	75	—	—
OAHl ≥ 3 RDI=2.1	91.7 (6.7)	92.9	83.3	70	92.8	16.5
OAHl ≥ 5 RDI=2.1	91.2 (6.4)	92.3	71.4	65	85.7	16.9
Children aged < 6 y (n=33)						
OAHl ≥ 1 RDI=3.35	76.9 (8.7)	73.1	71.4	78.7	90.4	58.5
OAHl ≥ 3 RDI=5.85	84.0 (7.2)	70.6	93.7	51.5	92.2	24.8
OAHl ≥ 5 RDI=5.85	88.7 (5.9)	78.6	89.5	42.4	84.6	15.0

Abbreviations: AUC, area under the receiver operating characteristic curve; NPP, negative posttest probability; OAHl, obstructive apnea-hypopnea index; PP, pretest probability or prevalence; PPP, positive posttest probability.

sensitivities, we should be cautious and view PSG as the gold standard diagnostic technique in routine clinical practice.^{5,6} The Spanish Working Group on Sleep²⁸ considers an OAHl between 1 and 3 in children to be diagnostic of SAHS, while according to the consensus reflected in the International Classification of Sleep Disorders, 1 or more respiratory events per hour of sleep in children is diagnostic of SAHS.²⁹

Few studies have investigated the role of respiratory polygraphy in the diagnosis of childhood SAHS and all of those have been carried out with a small number of patients, in selected populations, and without simultaneous comparison with PSG.¹⁰⁻¹³ The present study is the first in the literature reviewed to undertake a systematic study of the usefulness of respiratory polygraphy, and its reliability is due to the simultaneous performance of PSG and respiratory polygraphy. Our data showed a high rate of diagnostic agreement (84.9%) and a high correlation (89.4%) in the diagnosis of SAHS for an OAHl of 3 or more in PSG and an RDI of 3 or more in respiratory polygraphy. As the Bland-Altman plot shows, all but one of the values for the differences between OAHl and RDI fell within the 95% CI. Moreover, the distribution of values around the line corresponding to 0 on the axis representing the difference in means was random; in other words, respiratory polygraphy neither overestimated nor underestimated respiratory events compared to PSG. Agreement analysis using ROC curves showed respiratory polygraphy to have good validity with reference to PSG.

There is little agreement regarding what AHI value is clinically significant, and for this reason we chose the 3 OAHl values most often used in the literature. Calculating the ROC curves for each (≥ 1 , ≥ 3 , and ≥ 5), an RDI of ≥ 4.6 was found to be the cutoff with the greatest specificity and, consequently, with the smallest number of false positives. Even in children younger than 6 years, the specificity values obtained in our study showed a high level of protection against false positives.

It must be remembered that sleep architecture is preserved in children with SAHS, as was observed in our study (Table 1), and that a technique such as respiratory polygraphy can therefore provide a solution for the diagnosis of the disease, at least in theory. The main problem, however, is selecting the parameters to be measured.

Although we used a nasal pressure cannula in the PSG, a thermistor was used for the analysis and comparison of results of both diagnostic techniques. We observed, as have other authors,³⁰ that while the cannula tends to detect respiratory events like hypopneas more accurately than thermistors, there is a greater signal loss. The lack of oral airflow signal in a child is an important limitation that could be overcome with the use of an oronasal cannula in polygraphy recordings.

In the present study we did not develop a clinical model predictive of SAHS in children due to the high pretest probability of diagnosis in the population studied.

Our study has certain limitations: *a*) it was carried out on a selected population with a high probability of presenting SAHS; however, the aim of the study was to validate a simple system for diagnosing SAHS accurately

in children with clinical suspicion of the disease; *b*) carbon dioxide was not measured in the PSG, as it is a variable that is not usually recorded in respiratory polygraphy, and *c*) respiratory polygraphy was carried out in the sleep laboratory, although its greatest practical potential lies in home use; however, we believed it necessary first to compare respiratory polygraphy and PSG performed simultaneously.

Nevertheless, in hospital departments with experience in the diagnosis and treatment of sleep-disordered breathing, respiratory polygraphy can be used reliably in children to establish a diagnosis of SAHS and to make therapeutic decisions. This is of particular importance when PSG is unavailable.

The debate concerning the roles of respiratory polygraphy and PSG in children should not be treated as if a decision about which technique to use were to be made. Rather, both diagnostic techniques should be used to improve patient care, considering that an inaccurate evaluation of a child may lead to a failure to prescribe the appropriate treatment or to a decision to perform unnecessary surgery. The use of simplified tests for the diagnosis of SAHS, in children as well as in adults, should be undertaken within an appropriately coordinated system that includes respiratory polygraphy in the diagnostic algorithm. In cases of doubt as to diagnosis, PSG should be used.

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