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Original Article

Performance characteristics of the French version of the severity hierarchy score for paediatric sleep apnoea screening in clinical settings

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ABSTRACT

Background: Paediatric obstructive sleep apnoea syndrome (OSAS) is a highly prevalent condition carrying increased risk for impaired cognitive and cardiovascular function. The standard diagnosis consists of full-night polysomnography (PSG), but limited access to PSG leads to substantial under-diagnosis. The use of a validated and simple diagnostic screening tool to predict OSAS could prioritise night sleep recordings in children at risk of OSAS, and help in clinical decision-making.
Objective: This study aimed to prospectively assess the performance of the French version of the severity hierarchy score (SHS) in paediatric OSAS. This score consists of a discriminative subset of six respiratory items, and has already been validated in English for screening OSAS in the general paediatric population.
Methods: A total of 96 children (mean age 7.1 ± 2.4 years; BMI *z*-score: −0.03 ± 1.50) were recruited; they had been were referred to two academic sleep centres in France for the putative diagnosis of sleep-disordered breathing. The parents completed the SHS questionnaire prior to PSG. Sensitivity and specificity of the SHS for detecting moderate OSAS, defined by an apnoea–hypopnoea index (AHI) of ≥5/hours of total sleep time (TST), were assessed, and ROC analysis was performed.
Results: An SHS score of >2.75 exhibited an 82% sensitivity, 81% specificity, and 92% negative predictive value for detecting an AHI of ≥5/hour TST in the cohort.
Conclusion: The French version of the SHS emerged as favourably suited for the screening for OSAS in children.

1. Introduction

Obstructive sleep apnoea syndrome (OSAS) is a common disorder in otherwise healthy pre-school and school-age children, with an estimated prevalence of one to four percent in the general paediatric population [1]. The main pathophysiologic contributors include reduced airway size, increased ratio between the volume of the adenoids and tonsils, coupled with increased collapsibility of the upper airways, particularly during sleep [2]. Classically, OSAS in children is associated with delayed somatic growth and impaired school performance [3,4]. Adverse consequences of OSAS on endothelial function [5], systemic arterial blood pressure, cardiac geometry, and metabolic function have been described [6,7]. Adenotonsillectomy is generally effective in improving or normalizing the sleep-associated abnormalities [8] and is currently recommended as the first-line treatment for paediatric OSAS, particularly if the child is not obese and is less than seven years old [9].

In spite of its high prevalence, the potential associated risks for morbidities, and existence of effective treatment, OSAS remains largely under-diagnosed in the paediatric population. The main reason for such 'paradox' is that overnight polysomnography (PSG) – the gold-standard diagnostic test [10] – is not widely available. Despite the current guideline recommendations on the use of PSG,

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10% of children who snore and undergo adenotonsillectomy for OSAS are actually tested with a pre-operative PSG [11].

In this context, it would be desirable to develop a simple tool, such as a questionnaire-based approach, that would not only enquire about the main symptoms of OSAS, but would also provide a numerical score whose values correlate with the major diagnostic measurement derived from the PSG (ie, the apnoea–hypopnoea index (AHI). In 1984, Brouillette et al. [12] proposed the use of a questionnaire to evaluate the probability of OSAS in children, but the performance of this instrument fell short of the desirable operationally valid criteria that would enable its widespread use [13,14]. Indeed, clinical findings or symptoms, such as tonsillar size and snoring, reported by parents exhibit relatively high sensitivity but low specificity, while sleepiness symptoms, observed apnoea and difficulty breathing during sleep provide relatively high specificity but reduced sensitivity. Other proposed instruments, such as the respiratory score derived from the Paediatric Sleep Questionnaire [15] by Chervin et al. and the OSA questionnaire-18 by Franco and collaborators [16] have also been developed to help with the diagnosis; however, the length of the questionnaire or the complexity of calculation of the score have prevented their widespread use in the clinic. Other approaches combining symptoms and physical examination findings have been recently proposed, but await widespread trials [17]. Furthermore, none of the scores provided by these aforementioned questionnaires correlate with the value for PSG-derived AHI, and the scores only enable an estimate of the probability of having OSAS.

In 2012, Spruyt and Gozal developed a short questionnaire – the severity hierarchy score (SHS) – that was easy to use in the clinic [18]. This instrument consists of a hierarchic score of six questions and was validated in English in a general paediatric population. The relation between AHI and the score for this test is robust, with a score value of >2.72 reliably identifying children with an AHI >3/hours of total sleep time (TST), with a sensitivity of 60%, a specificity of 83%, and a negative predictive value of 93%, thereby eliminating the possibility of a potential diagnosis of OSAS with satisfactory precision.

For the present study, it was hypothesized that improved detection of children with more severe OSA, which is associated with increased risk for end-organ morbidity, would be desirable in a setting such as in France, in which access to sleep studies is relatively limited. The aim of the study was therefore to evaluate and validate the performance of the French version of the SHS [18] to diagnose at least moderate OSAS in a population of a higher pre-test probability (ie, habitually snoring children being evaluated pre-operatively in the Ear, Nose, and Throat department).

2. Patients and methods

2.1. Elaboration and characteristics of the French version of the severity hierarchy score questionnaire

Three separate groups of bilingual French clinicians translated the original questionnaire into French [18]. The three groups then fused the three proposed versions to form a single consensual version after universal agreement. This first French version was then translated back into English (counter-translation) by a French clinician whose mother tongue was English. The English back-translated version was then submitted to the author of the questionnaire (DG), who confirmed that the translation had not denatured the original version of the instrument. The original English version and the translated French version are shown in the Online Supplement. The French version will be referred to as the SHS questionnaire throughout this article.

2.2. Evaluation of the performance of the French version of the severity hierarchy score questionnaire

2.2.1. Study design

This was a prospective, non-randomised study. The study was carried out with the approval of the local ethics committee (CPP Ile-de-France V) regarding biomedical research.

2.2.2. Patients

Consecutive children with habitual snoring (defined as audible snoring reported by parents or caregivers >3/nights/week) but otherwise in good health were systematically included between July 2013 and October 2014. The children were all referred for possible adenotonsillectomy for suspected OSAS at two academic sleep centres (Saint-Antoine and Trousseau, Paris). The age range for inclusion was 3–13 years [19]. Exclusion criteria included the existence of a known chronic severe lung or cardiovascular disease, or the presence of a syndromic craniofacial malformation.

2.3. Data collection and polysomnographic recordings

An explanatory letter and the SHS questionnaire were provided to the children's parents, who completed it during the evening of the diagnostic PSG.

For the recording of night-time PSG, a CIDELEC polysomnograph was used (St Gemmes sur Loire, France) which enabled recording of several electrophysiological channels (three derivations of EEG; two electro-oculogram (EOG) channels; chin and leg electromyogram; and the following respiratory parameters: nasal air flow with a nasal cannula, respiratory effort using thoracic and abdominal belts, and sub-sternal chest pressure [20].

The polysomnograph was analysed by a sleep physician blinded to the SHS questionnaire according to the international recommendations of the American Academy of Sleep Medicine [10]. The SHS score was calculated according to the recommendations of Spruyt and Gozal [18] (see Appendix S1).

2.4. Definitions

Obstructive sleep apnoea syndrome was defined as an AHI of ≥5/hour TST, thereby establishing de facto two separate groups, namely: OSAS+ and OSAS− . The AHI cut-off was selected to define OSAS, as it indicates the inflexion point for the increase in cardiovascular and cognitive morbidities [5,21]. Obesity was defined by a BMI *z*-score of >1.65, according to the recommendations of the International Obesity Task Force [22].

2.5. Statistical analysis

Variables were expressed as percentages, mean ± SD, or median (IQR, interquartile range) values, as appropriate. Nominal variables were analysed with the Chi-squared test or Fischer's exact test. Quantitative variables were analysed with the non-paired *t* test, Mann–Whitney test or Spearman's correlation coefficient. A two-sided *p*-value <0.05 indicated statistical significance. Statistical and ROC analyses were performed with Statview 5.0. (SAS institute) and Stata (Stata/IC V 11.0), respectively.

3. Results

3.1. Study population

The study population consisted of 96 habitually snoring children – 29 girls and 67 boys – with a mean age of 7.1 ± 2.4 years (range 2.6–13.0). All parents who were approached agreed to participate

in the study. Their mean BMI *z*-score was −0.03 ± 1.50 (range −4.3–2.6; 95% CI −2.91, 2.97); 12.5% fulfilled BMI *z*-score criteria for obesity). The mean SHS score was 2.2 ± 1.0 (range 0–3.9; 95% CI 0.24, 4.16) and the mean AHI was 6.1 ± 11.4/hour TST (range 0–90/hour TST; 95% CI 0.82, 28.44). Of the 12 obese children (six boys), eight were OSAS−. There was no statistical relationship between the presence of obesity and inclusion in the OSAS+ group ($p = 0.74$).

3.2. Possibility of a centre effect

There were no significant differences between the two centres in terms of male/female distribution, age, BMI, AHI or SHS scores (Table 1).

3.3. Prevalence of obstructive sleep apnoea syndrome

According to definition, the prevalence of OSAS in the study sample was 29% (28/96 children). The characteristics of the children, according to the presence or absence of OSAS, are summarised in Table 2.

The OSAS+ group consisted of 21 boys (75%) and seven girls, compared with 46 boys (68%) and 22 girls in the OSAS− group. The values for the SHS, AHI, and oxygen desaturation index (ODI) were significantly higher in the OSAS+ group ($p < 0.0001$, Table 2). The distribution of AHI in the two groups is shown in Fig. 1. There were no significant differences between the OSAS+ and OSAS− groups in terms of male/female ratio, age, BMI, or total sleep duration (Table 2).

Correlation between apnoea hypopnoea index and severity hierarchy score

The relation between AHI values and corresponding SHS values is shown in Fig. 2 ($r = 0.514$, $p < 0.0001$), and ROC analysis of SHS in the prediction of OSAS is shown in Fig. 3 (AUC 0.87, $p < 0.001$). From the latter, an optimal SHS of 2.75 for detection of OSAS was retained (Table 3). Additional exploration of the ROC performance characteristics for the previously established optimal SHS, regarding detection of other AHI-based criteria of OSAS, revealed a reduced AUC for AHI >1.5/hour TST, with increased AUC values when severe OSAS criteria were applied (see Appendix S2).

Graphical determination of the optimal SHS value (the intersection point between the ROC curve and the diagonal) indicated a sensitivity of 82% and a specificity of 81%.

4. Discussion

This study showed that a simple six-question-based instrument, the SHS, enables delineation of a cut-off score value (ie, > 2.75), which allowed for remarkably accurate detection of OSAS in the present clinical referral cohort of children aged 3–13 years. The cohort included in this study was representative of the usual paediatric populations being evaluated in France for habitual snoring and being referred to a sleep centre before adenotonsillectomy. The present cohort was also comparable to those previously published, in terms of age and severity of OSAS [8,23,24].

Use of a lower AHI cut-off for OSAS, namely ≥1.5/hour TST [23], would lead to a prevalence of 68% in the present population. Notably, the performance of a SHS score in this context was less significant than when using an AHI cut-off of ≥5/hour TST and a SHS score of 2.75 (see Appendix S2). This high prevalence is unsurprising when taking into account the method of recruitment. In the study of Weatherly et al. [24], which employed similar inclusion criteria and age range, the prevalence of OSAS was 53%, and the mean AHI was 6.8 ± 8.4/hour TST; thus, the results of this study are remarkably similar to the present findings. In the population of 453 children included in the CHAT study (in which the definition of OSAS included an AHI ≥2/hour TST), the age range was again similar to the present study. In contrast to North-American studies [8], the children in the present study had an overall lower BMI, with 12.5% being obese, compared to 33% in the CHAT trial and 50.6% in the multicentre study of Bhattacharjee et al. [9]. No evidence was found for an association between obesity and OSAS, whereby the majority of children with abnormally high AHI values were not obese.

Although there are no precise cut-off AHI values demarcating between the presence or absence of neurocognitive dysfunction in children [5], a recent study [25] in a small group of apnoeic children aged 8–12 years, in whom working memory impairments were present, had a mean AHI of 5.6/hour TST. Similarly, an AHI of ≥5/hour TST reveals a much higher likelihood of cardiovascular morbidity [6]. On the basis of such considerations, in the present study, a cut-off AHI of ≥5/hour TST was selected as being of clinical significance rather than an epidemiologically defined AHI cut-off value of ≥1.5/hour TST. Such selection has been previously adopted, as illustrated by a recent study on the evolution of the prevalence of OSAS in a cohort of children or adolescents followed since birth [26]. Indeed, the authors decided not to use the epidemiological AHI cut-off, and also adopted an AHI of ≥5/hour TST as indicative of clinically relevant OSAS.

In a study evaluating the English version of the SHS, Kadmon et al. [27] also adopted the AHI threshold of ≥5/hour TST to classify the 85 subjects included in their study. However, the SHS cut-off value that discriminated children with an AHI of ≥5/hour TST from OSAS− children was 1.0, and exhibited a sensitivity of 83%, a specificity of 64%, PPV of 28%, NPV of 96%, and an AUC of 0.647. Compared to the present cohort, the study by Kadmon et al. included fewer children with an AHI >1.5/hour TST (34% vs 68% in the present study), and the children also had a lower AHI (3.4 ± 6.9/hour TST vs 6.1 ± 11.4/hour TST) and were slightly older (9.3 ± 3.5 years vs 7.1 ± 2.4 years). It is therefore possible that the differences in age, principally in disease severity, may account for the

Table 1
Comparison of the main characteristics of the study population in the two sleep centres.

	Centre 1 ($n = 59$)	Centre 2 ($n = 37$)	p
Sex (M/F)	40/19 (67.8/32.2%)	27/10 (73/27%)	0.07
Age (years)	7.4 ± 0.3	6.5 ± 0.3	0.07
BMI *z*-score	−0.001 ± 0.2	−0.089 ± 0.3	0.78
AHI (/hrTST)	7.2 ± 1.8	4.4 ± 0.7	0.24
SHS	2.1 ± 0.1	2.4 ± 0.2	0.28

Abbreviations: M, male; F, female; BMI, body mass index; AHI, apnoea–hypopnoea index; SHS, severity hierarchy score.

Table 2
Characteristics of the children according to the presence (apnoea–hypopnoea index ≥5/hrsTST) or absence of obstructive sleep apnoea syndrome.

	OSAS+ ($n = 28$)	OSAS− ($n = 68$)	p
Age (years)	6.5 ± 2.5	7.3 ± 2.3	0.06
BMI *z*-score	−0.2 ± 1.7	0.04 ± 1.4	0.59
SHS	3.1 ± 0.7	1.9 ± 0.9	<0.0001
AHI (/hrsTST)	16.5 ± 17.1	1.8 ± 1.3	<0.0001
ODI (/hrsTST)	12.5 ± 19.3	1.0 ± 1.3	<0.0001
TST (minutes)	524 ± 57.5	505.9 ± 71.1	0.44

Abbreviations: OSAS, obstructive sleep apnoea syndrome; SHS, severity hierarchy score; ODI, oxygen desaturation index; TST, total sleep time; AHI, apnoea–hypopnoea index; BMI, body mass index.

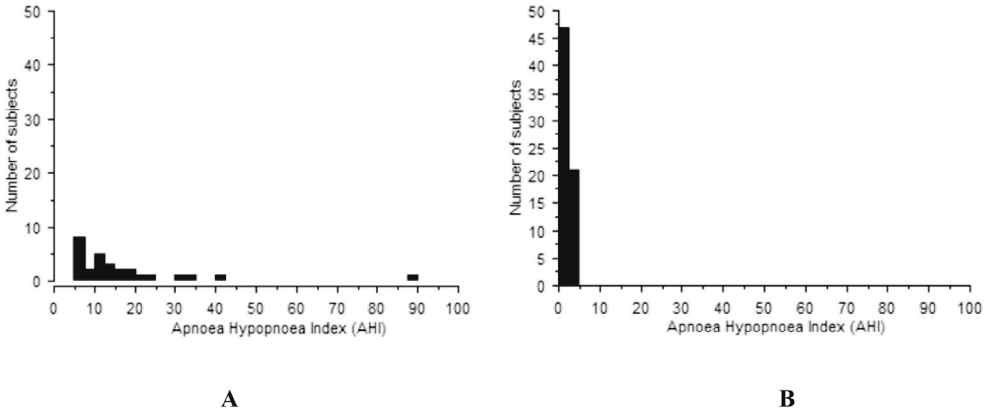

Fig. 1. Distribution of apnoea–hypopnoea index values as a function of the presence (A) or absence (B) of moderate obstructive sleep apnoea syndrome.

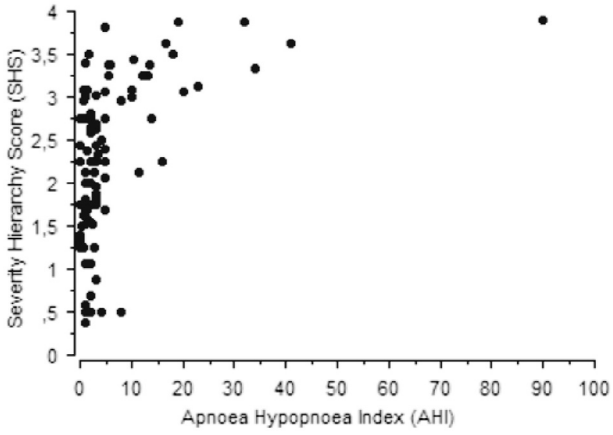

Fig. 2. Correlation between apnoea–hypopnoea index and severity hierarchy score. Graphical representation a scattergram plotting severity hierarchy score individual values against corresponding apnoea–hypopnoea index (/hrsTST) for the cohort of 92 children.

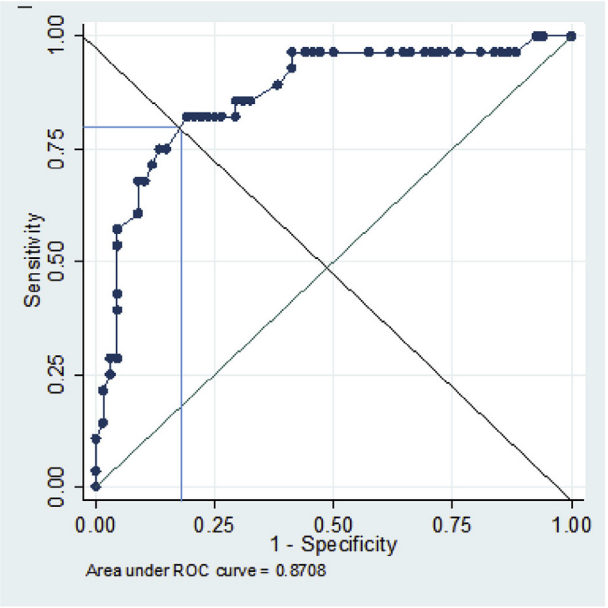

Fig. 3. ROC analysis of the performance of the severity hierarchy score criterion of 2.75 for detection of obstructive sleep apnoea syndrome (apnoea–hypopnoea index ≥5/hrsTST; see Appendix S2). Graphical determination of the optimal SHS value (the intersection point between the ROC curve and the diagonal) indicates a sensitivity of 82% and a specificity of 81%.

Table 3
Cut-off value for the severity hierarchy score yielding optimal prediction of obstructive sleep apnoea syndrome (AHI ≥ 5/hrTST).

SHS value	Sensitivity (%)	Specificity (%)	PPV (%)	PVN (%)
2.75	82.1	80.9	63.9	91.7
95% CI	74.4–89.8	73.0–88.8	54.3–73.5	86.2–97.2

Abbreviations: SHS, severity hierarchy score; PPV, positive predictive value; NPV, negative predictive value; CI, confidence intervals.

disparities in retained SHS cut-off values. Furthermore, cultural and genetic differences may have also contributed.

However, the present results were strikingly similar to those reported by Spruyt and Gozal [18]. Indeed, a cut-off value of 2.72 was derived for the SHS in their study, and enabled the best discrimination between children with OSAS and those without OSAS. In addition, both of these studies found positive correlations between the AHI values and SHS, as well as high AUC, thereby confirming the robustness of the current cut-off of 2.75 for the SHS to detect moderate OSAS in the present population. According to the sensitivity displayed by the SHS, screening in a large population emerges as a viable option for the instrument. However, even though the false positive rate was approximately 20%, since the major aim of the instrument is to define a valid tool for screening children at risk of OSAS, the sensitivity of the test should be prioritised over its specificity. The high NPV of 92% allows for effective elimination of the OSAS− cases when the score is <2.75, such that PSG (if available) and adenotonsillectomy without PSG can be safely avoided [28].

It is believed that there is no current validated clinical score that can be used in screening for OSAS. According to the authors, the respiratory score of the Paediatric Sleep Questionnaire [15], a 22-item questionnaire, is useful for research settings, but is not sufficiently reliable for individual patients. The OSA–18 questionnaire [29] has also been recognised as poorly adapted for routine clinical use. In 2014, Kadmon et al. took the concept of a diagnostic tool based on eight-items that were purely respiratory: the 'I'm Sleepy' questionnaire [27]. The performance of this score was illustrated by a sensitivity of 80% and a specificity of 50%, but this was an assessment a posteriori and not a prospective study, such that the tool did not allow a reliable prediction for any given individual child.

The present study had several limitations. First, a limited number of subjects were included, and the possibility that a larger number of subjects may have altered the cut-off value for the SHS cannot be excluded. A single PSG was performed and the possibility of inter-night variability of the AHI could exist, even though this is unlikely [30]. The present study was not carried out in the general

paediatric population, but despite the small sample size, the threshold value for the SHS was similar to that reported in the original study [18]. Two sleep centres participated in the assessment of the SHS; a centre effect was investigated and de facto eliminated, adding to the robustness of the findings. It should also be noted that the two centres used the same diagnostic recording system, which may have contributed to improved homogeneity of the PSG findings. This study was prospective and carried out under the usual conditions of daily clinical practice. The design was adapted to the problem of screening for OSAS in children who snore, irrespective of the initial route of referral. Future diffusion of this questionnaire to paediatricians, surgeons, ENT physicians, general practitioners, dental practitioners, and orthodontists, along with multicentre confirmation of the discriminant ability of the instrument, should allow for large scale and effective screening for OSAS, and rapidly identify those children who do not require further evaluation. In addition, in this cohort of 96 children who habitually snored, the French version of the SHS questionnaire on apnoeic severity enabled excellent discrimination of children with moderate OSAS, in whom potential surgical treatment would be indicated, using an SHS value of $\geq$2.75. Therefore, the questionnaire should prove a valuable adjunct for selection of children to be slated for PSG recordings for confirmation of the diagnosis.

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Conflict of interest

None.

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: http://dx.doi.org/10.1016/j.sleep.2016.01.021.

Appendix A. Supplementary material

Supplementary data to this article can be found online at doi:10.1016/j.sleep.2016.01.021.

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