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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

Xuân-Lan Nguyên [a, *], Pierre Lévy [b], Nicole Beydon [c], David Gozal [d], Bernard Fleury [a]

[a] Unité Sommeil et Fonction Respiratoire, Hôpital Saint-Antoine, Paris, France
[b] UPMC & Inserm UMR S 1136 (EPAR team), Département de Santé Publique, Hôpital Tenon, Paris, France
[c] Unité Fonctionnelle d'Exploration Fonctionnelle Respiratoire et du Sommeil, Hôpital Armand-Trousseau, Groupe Hospitalier Universitaire de l'Est Parisien, (APHP), Paris, France
[d] Sections of Pediatric Sleep Medicine and Pediatric Pulmonology, Department of Pediatrics, Pritzker School of Medicine, Biological Sciences Division, The University of Chicago, Chicago, IL, USA

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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,

* Corresponding author. Unité Sommeil et Fonction Respiratoire, Département de Physiologie Hôpital Saint-Antoine 184 rue du Faubourg Saint-Antoine, 75012 Paris, France.
E-mail address: nguyenxl@wanadoo.fr (X.-L. Nguyên).

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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