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Can a clinical score be used to screen for childhood OSAS?

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ABSTRACT

Pediatric obstructive sleep apnea syndrome (OSAS) is a frequent pathology (1-4% of the general population), often related to adenotonsillar hypertrophy. In France, however, access to polysomnography (PSG) is limited, leading to underdiagnosis. Using a simple, reliable diagnostic tool predictive of OSAS could prioritize prescription of night sleep recordings and help decision making for adenotonsillectomy. The aim of this study was to validate a French version of the sleep apnea Severity Hierarchy Score (SHS), already validated in English in the general population, for screening of childhood OSAS.

A prospective study included 86 children (aged 7.0 ± 2.4 years; BMI Z-score, –0.71 ± 1.51;) referred to 2 academic sleep centers, the Saint-Antoine and Trousseau hospitals (Paris, France) for assessment of sleep disordered breathing. The SHS questionnaire was filled out by the parents prior to overnight PSG. The sensitivity and specificity of the SHS were assessed according to various levels of OSAS severity.

A threshold of ≥2.75 on the SHS showed 92% sensitivity, 81% specificity and 96% negative predictive value for moderate to severe OSAS, defined by an apnea hypopnea index[3] of 5/hr in the study population.

KEY WORDS

Pediatric obstructive sleep apnea syndrome, diagnosis, polysomnography, questionnaire, validation

Obstructive sleep apnea syndrome (OSAS) consists in iterative pharyngeal occlusion due to imbalance between the mechanical load exerted on the pharynx and airway capacity to respond. It is a frequent pathology in children, especially of pre-school and school age, with a frequency peak at 6 years induced by an increased ratio between tonsillar volume and airway diameter. Airway obstruction by tonsillar hypertrophy is the most frequent cause of OSAS in children. Prevalence is 1% of the general pediatric population[6].

The clinical consequences of childhood sleep disordered breathing are diverse but specific, often consisting of retarded growth, enuresis, fatigue, somnolence or, in contrast, motor excitation. The most serious is neurocognitive impairment and learning difficulties[3,5]. Evidence of metabolic impact is increasing, with high blood pressure (notably deviation from normal day and

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night-time values and increased early morning peak) reported in children as being associated with moderately elevated apnea/hypopnea index (AHI) values of 5/h[1]. This 5/h threshold defines moderate to severe OSAS in the international classification of sleep disordered breathing. Data remain sparse on the relation between OSAS and endothelial function, although OSAS impact on the cardiovascular system can be demonstrated[4].

In a large majority of cases, it is airway obstruction by hypertrophic tonsils that underlies OSAS (figs 1 and 2) and, reassuringly, surgery is curative for 80% of children[7]. Surgical ablation of the tonsils and adenoid vegetation is thus still the main treatment for childhood OSAS, efficacy being especially sure in non-obese children aged less than 7 years[2].

Despite its high prevalence, however, OSAS is widely underdiagnosed in children due to the lack of typical clinical profile, symptoms being diverse and non-specific. Positive diagnosis requires polysomnography (PSG: i.e., complete recording of sleep and breathing), which is the reference examination but is expensive and complicated and available in few centers, largely accounting for failure to diagnose. For example, only 10% of children undergoing adenotonsillectomy, and therefore doubtless snorers, have preoperative PSG[10], although this is presently the best means of assessing the severity of obstruction, simple oximetry (nocturnal blood oxygen measurement) being contributive only when negative, and respiratory polygraphy (recording only cardiorespiratory parameters) tending to underestimate obstruction by underestimating AHI[9].

In this context, developing tools to facilitate screening for children with suspected OSAS and identifying high-risk cases so as to accelerate surgery becomes crucial.

David Gozal's team at the University of Chicago created a short questionnaire, validated in more than 1,000 children recruited via their schools, that can be implemented in routine practice; the score based on the subject's responses correlates strongly with AHI calculated from PSG[8]. The questionnaire comprises 6 questions, focusing on breathing (fig. 3). The final score takes account of differential weighting between questions: question 1

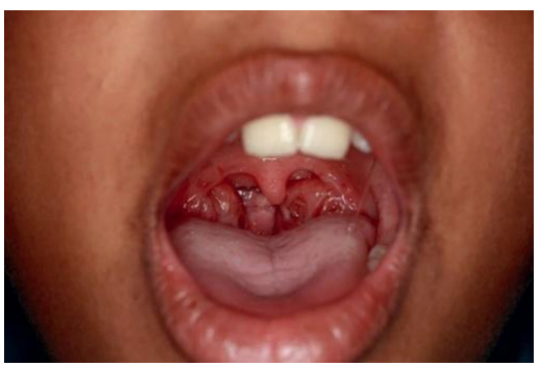

Figure 1
Obstructive tonsils in a 7 year-old boy.

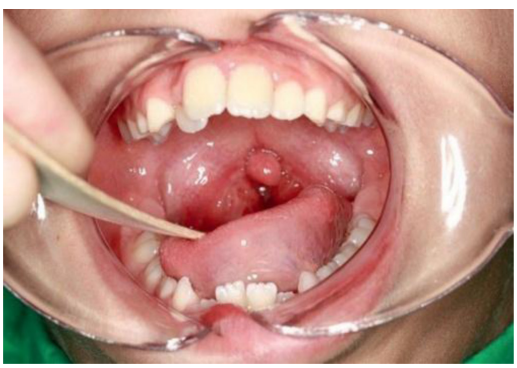

Figure 2
Obstructive tonsils in a 10 year-old girl.

In the last 6 months,

1. Have you had to help your sleeping child to start breathing again?
2. Does your child stop breathing while asleep?
3. Does your child have difficulty breathing while sleeping?
4. Has your child's breathing while asleep been a subject of concern for you?
5. How noisy is his/her snoring?
6. How often, does your child snore?

Responses to these questions are a score from 0 to 4 according to the frequency of the event

0 if "never"
1 if "rarely" (1 night per week)
2 if "occasionally" (2 nights per week)
3 if "frequently" (3 to 4 nights per week)
4 if "almost always" (more than 4 nights per week)

except for question 5, assessing snoring:

0: just perceptible or light snoring
1: moderate snoring
2: heavy snoring
3: very heavy snoring
4: extremely heavy snoring

Figure 3
Sleep apnea Severity Hierarchy Score questionnaire (Spruyt, Gozal CHEST 2012), to be filled out by the parents on the day of consultation.

("Have you ever shaken your child to get him or her to start breathing again?"), for example, is more contributive in diagnosing apnea than question 2 ("Does your child stop breathing during the night?"), which in turn is more contributive than questions 5 and 6, about snoring. The team thus demonstrated that this kind of questionnaire can reliably detect sleep disordered breathing in the general pediatric population. However, the score had not previously been assessed in a population consulting in a sleep center: i.e., with suspicion of OSAS based on presenting symptoms such as snoring or nocturnal breathing problems detected by the parents.

If such a diagnostically effective questionnaire could be used in routine practice, it would enable rapid screening of larger numbers of children, identifying those at high risk for OSAS who could then be quickly referred to an ENT

surgeon, and ruling out those without risk, who then would not have to undergo PSG, which would be reserved for doubtful cases. In children at high risk of OSAS, effectively identified from the questionnaire, initial PSG might not be needed, although the risk of recurrent or residual OSAS after surgery might indicate careful secondary surveillance[7]: i.e., regular postoperative clinical monitoring of the tonsils and vegetations, with repeat PSG in case of the slightest doubt. This attitude would allow for the need for extended pediatric screening while limiting public health expenditure.

A currently ongoing prospective study is assessing the performance of this sleep apnea Severity Hierarchy Score (SHS)[8] in screening for moderate to severe OSAS, defined by an AHI of 5/h, in a typical sleep center pediatric population consulting for suspected apnea/snoring. The 5/h threshold was chosen as being reasonably relevant from the clinical and cardiovascular points of view in the light of the literature on hypertension and childhood OSAS. Given the age range of children potentially concerned by the consequences of tonsillar hypertrophy, all 3-to-12 year-olds presumed to be in good health (excluding those with craniofacial deformity or serious respiratory pathology) were included. The SHS was filled out by the parents on the day of the diagnostic PSG. The PSG results were analyzed, following the international rules and regulations of the American Academy of Sleep Medicine, by an operator blind to the SHS results. All data were then entered in an Excel file for calculation of the final score.

A threshold SHS value was determined as having optimal sensitivity/specificity.

Between July 2013 and October 2014, 86 children referred for exploration of sleep disordered breathing consecutively underwent PSG at the Saint-Antoine and Trousseau hospitals in Paris, France. There were 26 girls and 60 boys, with a mean age of 7.0 ± 2.4 years, without obesity, mean percentage BMI being 52.9 ± 33.7%, or Z-score of –0.71 ± 1.51. Mean AHI was 5.9 ± 11.6/h, and mean SHS 2.2 ± 1.0. Fifty-six children (65%) had OSAS, defined as AHI ≥1.5/h, and 24 (27%) had moderate to severe OSAS, defined as AHI ≥5/h.

The 2.75 SHS threshold diagnosed OSAS with 92% sensitivity and 81% specificity, and 96% positive predictive value (i.e., 96% of children with scores <2.75 had AHI <5/h).

Analysis of the ROC curve confirmed that the 2.75 SHS threshold was the best compromise between sensitivity and specificity in this population, with an area under the curve of 0.9.

CONCLUSION

The sleep apnea Severity Hierarchy Score reliably screened for moderate to severe OSAS in 3-12 year-olds in the study population.

Routine implementation (systematic in case of snoring) could improve OSAS screening in community settings, preventing complications related to

non-treated childhood OSAS: retarded learning, cardiovascular complications.

It does not obviate the need for sleep PSG, but would improve the targeting and prioritization of examinations and reduce waiting lists. It could back up the ENT surgeon's opinion in case of certain OSAS and save time when the probability of OSAS is slight.

Conflict of interest: The authors have no conflict of interest to declare.

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