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Home respiratory polygraphy in obstructive sleep apnea syndrome in children: Comparison with a screening questionnaire

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ABSTRACT

Objectives: The prevalence of obstructive sleep apnea syndrome (OSAS) in children referred for sleep-disordered breathing reaches up to 59%. We aimed to test the adequacy of a questionnaire compared to home respiratory polygraphy (HRP), in 45 subjects (5–16 years-old), without maxillofacial malformations nor other comorbidities, presenting with symptoms compatible with OSAS.
Methods: All children passed a 12-items questionnaire (Obstructive Airway Child test: OACT) and the HRP. OSAS was classified in severity according to the apnea-hypopnea index (AHI).
Results: With HRP, 60% and 15% children were detected to have at least mild (AHI $\geq$1) and moderate (AHI >5) OSAS, respectively. The sensitivity of the questionnaire to detect mild and moderate OSAS was good (93% and 71%, respectively) but the specificity was very low (11% and 34%). However, an OACT score under 61 showed a very good negative predictive value for moderate and severe OSAS (87%). With the questionnaire, we could have avoided a complementary PSG or HRP in 25/45 (56%) of our subjects as in children with mild OSAS and without comorbidities only clinical observation is usually advised.
Conclusions: The OACT questionnaire has shown to be a good and quick instrument to exclude moderate and severe OSAS in our population of children without maxillofacial malformations. Indeed children scoring under 61 could avoid a constraining and expensive sleep exam. However, if the score is above this cut-off, the performance to recognize OSAS is low and the child's evaluation must be completed by a HRP or PSG.

1. Introduction

Obstructive sleep apnea syndrome (OSAS) affects about 2–4% of children aged between 2 and 8 years [24,30]. This respiratory disorder is characterized by recurrent episodes of upper airway obstruction during sleep, which are commonly associated with intermittent hypoxemia and sleep fragmentation [1]. In 2016 the European Respiratory Society Task Force published clinical practice guidelines for Diagnosis and Management of childhood OSAS [20]. Diagnosis of OSAS needs integration of symptom history, clinical and in particular ear, nose and throat (ENT) exam and polysomnographic or polygraphic findings.

Untreated OSAS may lead to significant neurocognitive and neurobehavioral dysfunctions [16,27], metabolic deregulations such as insulin resistance [14,29] and cardiovascular dysfunctions [31]. Symptoms for OSAS include frequent snoring, witnessed apneas, mouth breathing and fragmented sleep. Risk factors for OSAS are the presence of excess body weight with an almost 4-fold increased risk [30], adenotonsillar hypertrophy [4], which is also very frequent in obese children affecting 45% of them [13], craniofacial malformations, complex conditions such as Down syndrome, Prader-Willi syndrome and neuromuscular disorders.

Many questionnaires have been developed to predict OSAS in children; however, none of them were satisfactory until now [8,11,28,33].

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Abbreviations

AHI	Apnea-hypopnea index (episodes per hour)
BMI	Body mass index
ODI	Oxygen desaturation index
ENT	Ear, nose and throat
HRP	Home respiratory polygraphy
OACT	Obstructive Airway Child test
OSAS	Obstructive sleep apnea syndrome
PSG	Polysomnography
PSQ	Pediatric Sleep Questionnaire
ROC	Receiver operating characteristic

The traditional Pediatric Sleep Questionnaire (PSQ) has shown medium sensitivity and specificity (78 and 72%, respectively) to predict OSAS in a selected population of children addressed for adenotonsillectomy [10]. However, in 2012 a questionnaire, called Obstructive Airway Child test (OACT), has shown to be highly efficient to detect OSAS in concordance with polysomnography in children with craniofacial malformations [12]. Polysomnography (PSG) is considered the gold standard for the diagnosis of OSAS but in clinical practice the use of home respiratory polygraphy (HRP) has replaced progressively PSG because of its higher availability and cost effectiveness. Furthermore, it can be performed at home, in the child's natural environment [5,23,25,37]. In adults, it is considered as an appropriate alternative to PSG to diagnose OSAS [2,3,35], yet it has not yet been completely validated in children [23]. Nevertheless, some studies tend to show good results, especially with moderate and severe forms [2,20]. The diagnosis of OSAS is commonly based on the presence of sleep disordered breathing symptoms associated to an Apnea Hypopnea Index (AHI) $\geq$ 1 episodes per hour [36]. The European Respiratory Society Task Force recommended a more stringent cut-off with an AHI >5 to be used to determine children who would benefit from medical intervention [6,9,19] and an AHI of 1–5 in the presence of cardiovascular, neurologic or syndromic (Down or Prader-Willi syndromes) morbidities.

This study aimed to evaluate whether OACT questionnaire is suitable to screen for OSAS in a population of children without maxillofacial malformations suspected of OSAS and to compare it against home respiratory polygraphy (HRP).

2. Material and methods

2.1. Study design and subjects

This is a cross-sectional study held in the University Children's hospital of Geneva. We recruited a total of 45 subjects, aged 5–16 years, from the obesity, pulmonary and ENT clinics. In all subjects OSAS was suspected on the basis of snoring with or without witnessed nocturnal respiratory pauses, and/or multiple nights awaking, and/or diurnal sleepiness.

Children were excluded if presenting any of the following pathologies: 1) chronic pulmonary diseases (cystic fibrosis, interstitial pneumopathy, broncho-pulmonary disease, etc.); 2) acute asthma or uncontrolled asthma; 3) history of mechanical ventilation for lung diseases, tracheotomy or pneumonia during the previous year; 4) neuromuscular disease; 5) severe scoliosis; 6) craniofacial malformations; and 7) medications influencing sleep, blood pressure or breathing patterns.

The Mother and Child Ethics Committee of the University Hospitals of Geneva approved this study and a written informed consent was obtained from both parent and child (CER 12–221). This work has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki).

2.2. Measures

2.2.1. Anthropometrics

We assessed body weight (kg) in light clothes (panties and tee-shirt) and height (cm) without shoes. Body mass index (BMI) was calculated as weight/height squared ($kg \cdot m^{-2}$) and z-scores were derived using the World Health Organization references [17].

2.2.2. The obstructive airways child test (OACT) questionnaire

The OACT questionnaire [12] is composed of 12 questions. A score between 1 and 10 depending on the symptom's severity can be attributed to each question, with a minimal and maximal total score of 10 and 120, respectively. For this study the OACT questionnaire was translated into French by two independent translators (forward translation) and a consensus was developed from those translations by two independent pediatricians fluent both in French and English (consensus forward translation) (French version: see supplemental file). Then, this consensus version was back translated into English by two different independent translators who did not see the original questionnaire (back translation).

All caregivers of the patients had to fill in the French version of the OACT questionnaire under the research nurse supervision as to check for completeness. There was no missing data. In function of the total OACT score obtained, OSAS was diagnosed as mild (score 31–60), moderate (score 61–90) or severe (score 91–120).

2.2.3. Home respiratory polygraphy (HRP)

The HRP was performed in all subjects using the Embla® Embletta® GOLD portable sleep system (ResMed). During one night of sleep in the child's home setting, we recorded nasal airflow (nasal canula pressure transducer), respiratory movements and their sum (dual thoracic and abdominal respiratory inductance plethysmography belts), body position (system position), snore (nasal pressure transducer), oxygen saturation SpO2 beat-to-beat and average by pulse oximetry and pulse rate by oximeter. A research nurse trained in pediatric sleep techniques explained to the child and the caregiver how to position, replace and remove the sensors. The child was equipped by the nurse in hospital with the recording system, belts, oximeter and nasal canula. The records were then performed at home without supervision as already published [26]. The child and/or his caregivers were asked to fill in a diary indicating times of falling asleep, awaking and any incidents during the night, like waking up or going to the toilet. The polygraphic recording was analyzed by using the RemLogic-E™ software and manually scored by two trained MDs who were blind to the questionnaire result.

Apnea was defined, according to pediatric scoring rules of the American Academy of Sleep Medicine 2012 [6], as a drop in the peak signal excursion of the nasal flow trace or belt sum trace (sum of thoracic and abdominal belts' traces) by $\geq$ 90% of the pre-event baseline for at least the time equivalent to two respiratory cycles. When respiratory efforts were maintained it was scored as an obstructive apnea. When inspiratory efforts were absent and associated with $\geq$3% oxygen desaturation, it was scored as a central apnea. Hypopnea was defined as a decrease of $\geq$30% in the amplitude of nasal flow trace or belt sum trace during the time equivalent to two respiratory cycles, associated with a drop in oxygen saturation of $\geq$3%. The Apnea Hypopnea Index (AHI) was defined as the total number of respiratory events (apneas plus hypopneas) divided by the total analyzed study time in hours. The mean oxygen saturation was recorded and the number of oxygen desaturations $\geq$3% divided by the total analyzed study time in hours was defined as the oxygen desaturation index (ODI). HRP tests were considered un-interpretable if the key signal (oxygen saturation) was absent and/or no reliable airflow and belt sum trace signals, and if analyzed time was less than 300 min. In those situations the subjects were excluded from the analysis.

We used the ERS OSAS-Definition 2 [20] to diagnose and classify OSAS: presence of symptoms of sleep disordered breathing and AHI: <1:

no OSAS; 1–5: mild OSAS; >5–10: moderate OSAS; and >10 episodes per hour severe OSAS [7,18,34]. As for mild OSAS only clinical observation is recommended, and no treatment proposed, especially in children without comorbidities, we used an AHI >5 as cut-off value for a further data analysis. Indeed, children with moderate and severe OSAS are unlikely to resolve OSAS spontaneously and would benefit from a therapeutic management that may include several options ranging from weight loss to surgical interventions and non-invasive ventilation [9, 19].

2.3. Statistical analysis

Statistical analyses were performed using the SPSS software 18.0 (Chicago, IL). Descriptive analysis was performed using frequency distributions for the qualitative variables and mean and standard deviation (SD) for the quantitative ones. We used independent Student's t-test, Chi-2 and bivariate Pearson correlation when appropriate. Differences were considered significant if $p < 0.05$. Diagnostic efficiency receiver operating characteristic (ROC) curve was calculated and the following cut-off point was used for the validity analyses: AHI >5. Sensitivity, specificity, as well as positive and negative predictive values were obtained to the diagnosis of moderate and severe OSAS. The Cronbach's alpha coefficient were calculated to estimate the internal consistency of the questionnaire for our sample size, and was considered acceptable if > 0.7.

3. Results

3.1. Patients' characteristics

We included a total of 48 subjects, but three had to be excluded from the analyses due to non-interpretable HRP. Therefore, we present the results of the 45 subjects who had valid data for both the OACT questionnaire and HRP test.

Characteristics of the subjects are presented in Table 1.

Table 1
Characteristics of subjects. Results are expressed as mean and SD.

	Subjects
	N = 45
Gender female: n (%)	16 (35.6)
Age (years)	8.9 ± 3.5
Age range (years)	5–16.3
Anthropometrics	
Weight (kg)	45.1 ± 29.6
Height (cm)	135.0 ± 21.6
BMI ($kg.cm^{-2}$)	22.3 ± 8.0
BMI z-score	1.7 ± 2.2
Normal weight (n = 17) BMI z-score	−0.6 ± 0.9
Overweight (n = 4) BMI z-score	1.5 ± 0.3
Obese (n = 24) BMI z-score	3.4 ± 1.1
OACT questionnaire	
Total Score (range)	12–89
Total Score (mean)	56.1 ± 19.3
No OSA (<31): n (%)	4 (9)
Mild OSA (31–60): n (%)	21 (46.6)
Moderate OSA (61–90): n (%)	20 (44.4)
Severe OSA (91–120): n (%)	0
Home Respiratory Polygraphy	
Mean oxygen saturation (%)	96.6 ± 1.1
ODI (events/h)	5.2 ± 9.1
AHI (events/h)	3.0 ± 5.9
OSA severity:	
No OSA (AHI <1): n (%)	18 (40)
Mild OSA (AHI: 1–5): n (%)	20 (44.4)
Moderate OSA (AHI: >5–10): n (%)	3 (6.7)
Severe OSA (AHI: >10): n (%)	4 (8.9)

Abbreviations: BMI: body mass index; OSA: obstructive sleep apnea; ODI: oxygen desaturation index; AHI: apnea/hypopnea index.

24/45 (53.3%) were obese (BMI z-score >2), 4/45 (9%) overweight (BMI z-score 1–2) and 17/45 (37.7%) had normal weight.

3.2. OACT questionnaire

41/45 (91%) had scores compatible with mild (n = 21) and moderate (n = 20) OSAS, but none for severe OSAS (Table 1). The Cronbach's alpha coefficient calculated with our sample was acceptable (0.77).

3.3. Home respiratory polygraphy

27/45 (60%) subjects were diagnosed to have OSAS (AHI ≥1), with 20/45 mild (AHI 1–5), 3/45 moderate (AHI 5–10), and 4/45 severe OSAS (AHI: >10). Total for moderate and severe 15% (Table 1).

3.4. Correlation between OACT scores and HRP indices

The total OACT score was positively but poorly correlated to the AHI ($r = 0.317$, $p = 0.034$; Fig. 1) and the ODI ($r = 0.315$, $p = 0.035$).

Neither the OACT nor the HRP values were related to the patients' age or the BMI z-scores, except for the mean oxygen saturation which was inversely correlated to the BMI z-score ($r = -0.365$, $p = 0.014$).

Table 2 presents the analysis of the concordance between the OSAS severities obtained with the questionnaire and the HRP test. It shows that there is no significant correlation between both evaluations. Only mild OSAS on questionnaire seemed to be related to mild OSAS on HRP in 12/20 (60%) children.

However, mean OACT score was significantly higher in children with an AHI >5 (Fig. 2). With an AHI cut-off of >5 the AUC (area under the curves) was of 0.750.

The sensitivity and specificity of the OACT questionnaire compared to HRP for the cut-off of AHI >5/h is presented in Table 3. With this cut-off of reference, the questionnaire showed good sensitivity and negative predictive value.

Finally, we looked at the questions individually to see if any one of them was independently correlated to OSAS severity and found a significant result for only 2 of them (Q4: $p = 0.013$; and Q5: $p = 0.048$). Question four was related to the intensity of snoring and question five to the visualization of apnea by the caregiver.

4. Discussion

The results of our study show that the OACT questionnaire is sufficiently strong to exclude moderate and severe OSAS in our population when scoring under 61. However, higher scores were not well correlated to AHI, overestimating OSAS, underlying the need for complementary sleep testing in those children.

Our subjects were suspected of OSAS because of snoring, visualization of apnea by parents or diurnal fatigue, without maxillofacial malformations or other comorbidities. The proportion of OSAS in our selected population was quite similar to the one found in other studies, ranging from 51% to 59% [21,22,32]. Indeed, we found that 60% of them suffer of at least mild OSAS according to AHI. In our study we focused on children presenting moderate and severe OSAS, which represented 15% of our population.

We looked for the concordance between OSAS severity diagnosed either by HRP or OACT questionnaire. In that group, we found that the correlation between AHI and OACT score was significant, but relatively poor. Indeed, the questionnaire score is dependent on the parent's subjective appreciation, which may overestimate the importance of clinical symptoms. The only questions that seemed to be independently correlated to the OSAS severity were the snoring intensity and the visualization of apnea by the parents, which was already described in a preceding study [33].

As in clinical practice we are interested to detect primarily children who need treatment interventions, we analyzed more specifically the

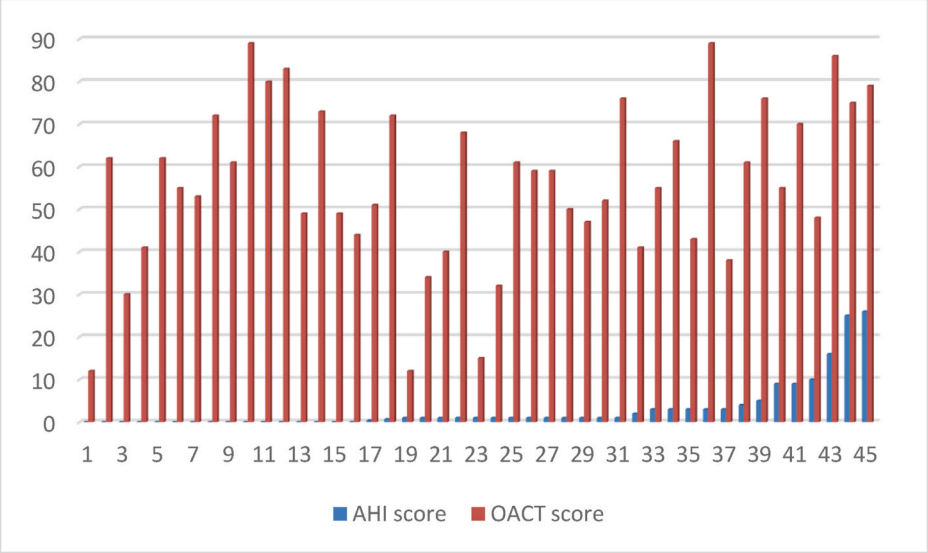

Fig. 1. Correlation between individual (n = 45) OACT total score and AHI. Abbreviations: AHI: apnea/hypopnea index; OACT: obstructive airways child test.

Table 2
OSAS severity classifications according to OACT questionnaire and HRP.

	AHI			
	Normal (AHI: <1)	Mild (AHI: 1–5)	Moderate (AHI: >5–10)	Severe (AHI: >10)
OACT score				
Normal (<31)	2	2	0	0
Mild (31–60)	7	12	1	1
Moderate (61–90)	9	6	2	3
Severe (91–120)	0	0	0	0
Total	**18**	**20**	**3**	**4**

Abbreviations: OSAS: obstructive sleep apnea syndrome; AHI: apnea/hypopnea index.

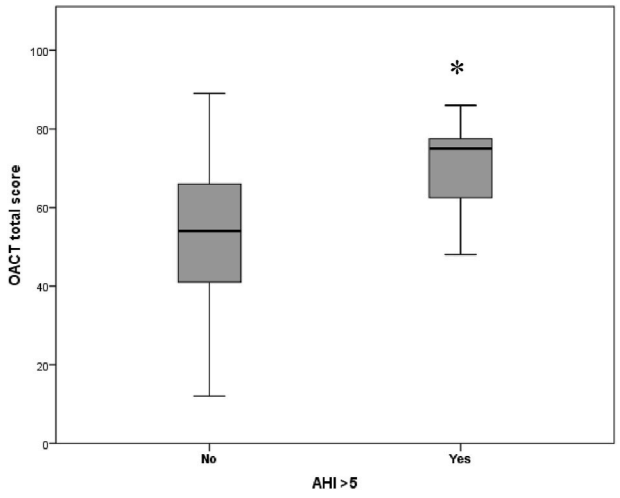

Fig. 2. Relationship between mean OACT score and AHI cut-off >5. Abbreviations: AHI: apnea/hypopnea index; OACT: obstructive airways child test.

ability of the questionnaire to detect children with moderate and severe OSAS (AHI >5), as done previously [28]. In children with an AHI >5, the mean OACT score was significantly higher (Fig. 2), however, children with mild OSAS could also have high scores (Fig. 1). Indeed, looking at

Table 3
Diagnostic validity for OACT questionnaires compared to HRP.

	Cut-off reference AHI >5
	OACT
	≥61
Sensitivity, %	71.4
Specificity, %	34.2
Positive predictive value, %	25
Negative predictive value, %	86.7

Abbreviations: AHI: apnea/hypopnea index; OACT: obstructive airways child test.

the moderate and severe OSAS according to the OACT questionnaire (score 61–120), the sensitivity was acceptable (71%), but the specificity was low (34%) (Table 3). Therefore, we may conclude that a questionnaire score above 61 is not sufficient to diagnose moderate and severe OSA, and those children should perform a sleep exam.

More interestingly, our study showed that a score under 61 can reasonably exclude moderate and severe OSAS, as the negative predictive values was of almost 87%. Taking into account this analysis, we could have avoided a complementary PSG or HRP in 25/45 (56%) of our subjects. Our finding is in line with a previous study published in 2018, which evaluated in 420 subjects without comorbidities a 5 items questionnaire to screen for OSAS. Their negative predictive value for moderate and severe OSAS was between 81 and 88% [33]. They concluded similarly that a questionnaire may be used as a screening tool to exclude moderate and severe OSAS and therefore may enable to reduce the number of investigations in those children, but cannot diagnose correctly OSAS severity.

The discrepancy of our results compared to those of the team who developed the questionnaire [12], may be explained by the very different population of both studies. Indeed, they tested their questionnaire on 35 children with maxillofacial malformations who achieved higher PSG indices and questionnaire scores. Another potential explanation would be that clinical symptoms were either not well reported, depending on subjectivity or the questions were not sufficiently pertinent. We can therefore assume that the OACT questionnaire is better to exclude moderate or severe OSAS and cannot be generalized to other pediatric populations, probably because of distinct clinical pattern of OSAS. Another study also demonstrated that a questionnaire can perform differently depending of the studied population, for example

with or without comorbidities [28].

Many questionnaires have been used to detect OSAS compared with polysomnography. In one study, the authors performed polysomnography on 85 children referred for sleep study, with the following AHI cut-off values to diagnosis OSAS: AHI $\geq 1.5 < 5$ for mild OSAS, AHI $\geq 5 < 10$ for moderate OSAS and AHI ≥ 10 as severe OSAS which is quite similar to our study [21]. The evaluated questionnaire was composed of 6 questions following a "Likert-type" response scale. The predictability of their questionnaire for mild OSAS was also low (20%), but it had a much higher sensitivity and specificity for moderate and severe OSAS (sensitivity 83%, specificity 64%) than in our study. However, their ROC curve result was lower than ours (AUC = 0.647 vs. 0.750). These authors proposed that discrimination of moderate to severe from mild OSAS was indeed very important as it will enable to treat children at increased clinical risk. They emphasized the need for a short and simple questionnaire like their 6-item sample. Another study, performed in 105 children scheduled for adenotonsillectomy, compared the Pediatric Sleep Questionnaire (PSQ) results to polysomnography [10]. They found a sensitivity of 78% and a specificity of 72% to detect OSAS (defined as obstructive apnea index ≥ 1). However, this questionnaire was constituted of 22-items and took 20–30 min to complete, making it more difficult to use in daily clinical practice.

Our study showed some limitations. First our sample size was quite small, as the one used to validate the OACT questionnaire. Furthermore, we used HRP instead of PSG, as recent studies concluded that HRP was a useful alternative to polysomnography for diagnosing OSAS in children [2,15].

5. Conclusions

In conclusion, we demonstrated in our population of children without maxillofacial malformations or comorbidities that the OACT questionnaire is a good and quick instrument to determine which child would need a complementary sleep testing. Indeed children detected without or with mild OSAS by the questionnaire (score under 61), were well identified not to have moderate and severe OSAS (AHI >5) with HRP. Now, it would be very useful to develop a more specific questionnaire to confirm moderate and severe OSAS, as to reduce the numbers of PSG or HRP also in those children. Finally, the OACT questionnaire would be worthwhile to be validated in a more divers and large population.

Declaration of competing interest

None.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ijporl.2021.110635.

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Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.

Informed consent

Informed consent was obtained from all individual participants included in the study.

Author's contribution

AM conceptualized the study, coordinated the study, performed the statistical analysis, drafted the initial manuscript, and approved the final manuscript as submitted. RC participated in the study design, drafted the initial manuscript and approved the final manuscript as submitted. CGS: participated in the study design, collected the data and approved the final manuscript as submitted. MB, HCV and CB: participated in the study design and approved the final manuscript as submitted.

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