Sleep and Breathing (2022) 26:763–769
https://doi.org/10.1007/s11325-021-02471-4

SLEEP BREATHING PHYSIOLOGY AND DISORDERS • ORIGINAL ARTICLE

Accuracy of the sleep clinical record for the diagnosis of pediatric moderate-to-severe obstructive sleep apnea syndrome

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Received: 22 April 2021 / Revised: 31 July 2021 / Accepted: 3 August 2021 / Published online: 8 August 2021

Abstract

Purpose The sleep clinical record (SCR) has been used to diagnose obstructive sleep apnea syndrome (OSAS) in children when access to polysomnography (PSG) is limited. Our aim was to determine the best SCR score that could facilitate diagnosis of moderate-to-severe OSAS in children with snoring.

Methods Healthy children with history of snoring, who were referred for PSG, were prospectively recruited. The SCR score was calculated. Receiver operating characteristic curves (ROCs) were plotted to determine the area under curve (AUC), and the optimum SCR cutoff value was determined using the Youden index (*J*).

Results Two hundred and seventy-three children were recruited (mean age 6.3 ± 2.5 years; median obstructive apnea–hypopnea index 1.5 episodes/h; range 0–61.1). The mean SCR score was 6.9 ± 3.6. Forty-six children had moderate-to-severe OSAS. Subjects with moderate-to-severe OSAS had a significantly higher mean SCR score (10.2 ± 2.9) than those with mild OSAS (6.2 ± 3.3; $P < 0.001$). Based on the plotted ROC, the AUC was 0.811 (95% confidence interval: 0.747–0.876; $P < 0.001$). Calculation of *J*, based on its ROC coordinates, indicated that the optimum cutoff SCR score to predict moderate-to-severe OSAS was 8.25, corresponding to a sensitivity of 83% and a specificity of 70%.

Conclusion Among children with history of snoring, an SCR score above 8.25 can identify those with moderate-to-severe OSAS.

Keywords Children · OSAS · Pediatric sleep questionnaire · Polysomnography

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Introduction

Obstructive sleep-disordered breathing (SDB) consists of several clinical entities ranging from the mildest disorder, which is the primary snoring (PS), to obstructive sleep apnea syndrome (OSAS). Increased upper airway resistance and collapsibility of the pharynx during sleep predispose to recurrent apneas and hypopneas [1]. Additionally, OSAS affects 1 to 4% of children worldwide [1]. Nocturnal polysomnography (PSG) represents the gold standard test for diagnosing and measuring severity of OSAS [1]. PSG is a high-cost test that requires clinical expertise and monitoring in the sleep laboratory. Early adenotonsillectomy (AT) is associated with favorable outcomes [2]. Due to the limited number of pediatric sleep laboratories and the high cost of the test, fewer than 10% of children with snoring who are scheduled for adenotonsillectomy undergo PSG [3]. As a diagnostic alternative, nocturnal pulse oximetry provides ease of both application and interpretation [4]. Moreover, it

has a lower cost compared to PSG. Despite its advantages, the requirement of night recording should not be overlooked. The difficulty of diagnosing all children with SDB by PSG or nocturnal pulse oximetry has resulted in efforts to establish clinical scoring tools that can accurately estimate the risk of being diagnosed with OSAS.

Several questionnaires have been devised to identify children with possible OSAS [5–8]. Questionnaires structured upon the disorder symptoms are helpful screening tools, but they cannot replace PSG [1]. To improve the diagnostic ability of clinical questionnaires, researchers have tried to integrate findings from the physical examination. Villa et al. proposed such a questionnaire, which is known as the "sleep clinical record" (SCR) [7]. The SCR contains three elements: physical examination findings, the patients' subjective symptoms, and presence of inattention and hyperactivity [7]. An SCR score ≥ 6.5 in children with snoring provides a sensitivity of 96%, specificity of 67%, a positive likelihood ratio of 2.91, a negative likelihood ratio of 0.06, and accuracy of 88% for predicting OSAS (AHI ≥ 1 episode/h) [7]. In a subsequent study, Villa et al. reported that both the sensitivity and positive predictive value (PPV) of the SCR for diagnosing OSAS were 91%, whereas specificity and negative predictive value (NPV) were 40% [9]. In a European Respiratory Society statement, pediatric sleep questionnaire and the SCR were considered acceptable solutions for diagnosing and determining the severity of OSAS if PSG is not available [1].

Treatment of OSAS is especially beneficial when apnea–hypopnea index (AHI) exceeds 5 episodes/h [1]. Consequently, it is essential to know whether a child with SDB symptoms has mild or moderate-to-severe OSAS. An SCR score ≥ 6.5 in children with snoring has been evaluated for detecting children with mild OSAS (AHI ≥ 1 episodes/h), but not subjects with moderate-to-severe OSAS [7, 9]. Thus, this study aimed to estimate the worth of the SCR in diagnosing children with clinically suspected moderate or severe OSAS and to determine the best cutoff SCR score for this purpose.

Methods

Participants and study protocol

The Larissa University Hospital Scientific Council gave its approval to this research (9678/02.27.2018). All participants' parents or legal guardians gave their informed consent. Consecutive children between 2 and 16 years old who had snoring and were referred for PSG were prospectively recruited. The following were the exclusion criteria for study participation: (1) signs of an acute respiratory tract infection; (2) prior AT or any other treatment interventions for OSAS; (3) a history of craniofacial abnormalities, neuromuscular, neurocutaneous, or genetic diseases, congenital heart disease, or chronic lung disease.

Initial clinical evaluation and SCR completion

Physical examination findings, the patients' subjective symptoms, and features of inattention and hyperactivity are the three components of the SCR. The first section includes the clinical examination of the nose, oropharynx, dental, and skeletal occlusion. The second section documents the patient's SDB subjective symptoms as measured by the Brouillette score [5]. The third part uses a disorder rating scale completed by the parents to identify symptoms of inattention and hyperactivity [10].

Children who were scheduled for PSG arrived late in the afternoon at the sleep laboratory. After being interviewed, they were submitted to a focused physical examination by one of the investigators who were trained in the completion of SCR. Somatometrics were recorded. Visual evaluation of the oropharynx was completed, and tonsillar size was graded from 1 + to 4 + and noted when the size was greater than 2 + [11].

PSG

Laboratory PSG confirmed the diagnosis of OSAS. A computerized Alice 5 system (Healthdyne; Marietta, Georgia) was used to perform an overnight PSG in the Sleep Disorders Laboratory. The following parameters were recorded: electroencephalogram, electrooculogram, submental and tibial electromyogram, body position, electrocardiogram, thoracic and abdominal wall motion (piezoelectric transducers), oronasal airflow (oronasal thermistor and nasal pressure transducer), and oxygen saturation of hemoglobin. A physician trained in sleep medicine scored the PSG recording and analyzed the results. The American Academy of Sleep Medicine manual was used to score PSG [12]. OSAS severity was classified based on the AHI and children with AHI > 5 episodes/h were diagnosed with moderate-to-severe OSAS. The sleep physician who scored the sleep studies was blinded to the SCR results.

Data analysis

Statistical analysis was carried out using the chi-square test for nominal variables and *t*-test for independent samples and scale variables. Receiver operating characteristic curves (ROCs) were plotted to determine the area under curve (AUC). Subsequently, the optimum SCR score cutoff value was obtained accordingly by estimating the Youden index (*J*) [13]. *J* is formally defined as $\max_c$ {Sensitivity (c) + Specificity (c) − 1}, where "max" is the maximum of

the sum of [Sensitivity + Specificity − 1], achievable for *c*, the optimal cutoff value for the biomarker under scrutiny. The biological significance of *J* is that it represents the best possible combination of sensitivity (*c*) and specificity (*c*). These analyses were conducted via the SPSS software (version 26.0; SPPS Inc., Chicago, IL).

Results

A total of 290 PSGs were completed during the study period, and 273 of them were performed on children aged 2–16 years who did not meet any of the exclusion criteria. Ten children were excluded due to a previous AT and five children because they had a history of craniofacial abnormalities, and neuromuscular, neurocutaneous, or genetic diseases. Additionally, the SCR was not completed for two children.

The mean age of the 273 participating children was 6.3 ± 2.5 years, with 115 of them being female. The median obstructive AHI was 1.5 episodes/h with range 0–61.1 episodes/h. Forty-six (17%) out of 273 children met the criteria for moderate or severe OSAS (AHI > 5 episodes/h), while 227 (83%) children met the criteria for mild OSAS (AHI 1–5 episodes/h) or PS (AHI < 1 episodes/h). Characteristics of children according to the severity of SDB are shown in Table 1.

Values of PSG indices and SCR score are summarized in Table 2. Subjects with mild OSAS or PS did not differ from children with moderate-to-severe OSAS concerning age, gender, and percentile of BMI *z* score (Table 1). The moderate-to-severe OSAS group exhibited nasal septum

Table 2 Polysomnography indices and SCR score

	AH ≤ 5 episodes/h	AHI > 5 episodes/h	*P*-value
Subjects	227 (83)	46 (17)	
AHI, episodes/h	1.4 ± 1.3	11.2 ± 8.5	< 0.001
DI, episodes/h	2.5 ± 2.6	18.7 ± 23.1	< 0.001
Minimum oxygen saturation, %	90.8 ± 3.5	85.2 ± 4.9	< 0.001
RAI, episodes/h	0.7 ± 0.8	3.4 ± 4.8	< 0.001
SCR score	6.2 ± 3.3	10.2 ± 2.9	< 0.001

Data are presented as *n* (%) or mean ± SD, *P*-value calculated by *t*-test. *AHI*, apnea–hypopnea index; *DI*, oxygen desaturation (≥ 3%) index; *RAI*, respiratory arousal index; *SCR*, sleep clinical record

Table 1 Characteristics of children according to the severity of SDB

	AHI ≤ 5 episodes/h	AHI > 5 episodes/h	*P*-value
Subjects	227 (83)	46 (17)	
Male	134 (58)	22 (52)	NS
Age, years	6.3 ± 2.4	6.4 ± 3.3	NS
BMI *z* score	0.33 ± 1.57	0.54 ± 1.63	NS
Nasal septum deviation	11 (5)	12 (29)	< 0.001
Nasal cartilage hypotonia	28 (11)	20 (44)	< 0.001
Tonsillar hypertrophy	143 (63)	38 (83)	< 0.05
Friedman palate position	64 (28)	29 (63)	< 0.001
Orbicular muscle hypotonia	29 (13)	23 (50)	< 0.001
Nasal airway patency	116 (51)	37 (80)	< 0.001
Habitual nasal obstruction	176 (78)	43 (94)	< 0.05
Open bite	8 (4)	0 (0)	NS
Deep bite	13 (6)	5 (11)	NS
Cross bite	16 (7)	6 (13)	NS
Overjet	10 (4)	1 (2)	NS
Narrow palate	103 (45)	41 (89)	< 0.001
Normal phenotype	160 (71)	14 (30)	< 0.001
Adenoid phenotype	29 (13)	20 (43)	< 0.001
Adult phenotype	37 (16)	11 (24)	NS
Witnessed apneas	174 (77)	44 (96)	< 0.05
Habitual snoring	221 (97)	46 (100)	NS
Positive Brouillette score	163 (72)	44 (96)	< 0.001
Positive for ADHD	56 (25)	16 (35)	< 0.05

Data are presented as *n* (%) or mean ± SD, *P*-value calculated by *t*-test, and others calculated by chi-squared test. *NS*, not significant; *AHI*, apnea–hypopnea index

deviation, nasal cartilage hypotonia, tonsillar hypertrophy, orbicular muscle hypotonia, habitual nasal obstruction, higher Friedman tongue position score, and nasal obstruction in contrast to subjects with mild OSAS or PS (Table 1). Using the originally reported cutoff value of 6.25 to set the SCR as positive, 46 out of 153 children with positive SCR had moderate-to-severe OSAS. All children with negative SCR had mild OSAS or PS. Subjects with moderate-to-severe OSAS had a significantly higher mean score in SCR (10.23 ± 2.86) than those with mild OSAS (6.20 ± 3.34; $P < 0.001$). The SCR had a sensitivity of 100% and a specificity of 53% for diagnosing children with moderate-to-severe OSAS. The PPV was 30% and the NPV was 100%.

We then calculated the optimal cutoff value of the SCR to predict subjects with moderate-to-severe OSAS. Based on the plotted ROC (Fig. 1), the AUC was determined to be 0.811 (95% confidence interval: 0.747–0.876; P-value < 0.001). Calculation of J, based on its ROC coordinates, indicated that the optimum value for detecting moderate-to-severe OSAS was 8.25, corresponding to a sensitivity of 83% and a specificity of 70%. The PPV was 36% and the NPV 95%. Based on the SCR score > 8.25, 83% of the children with OSAS were properly diagnosed.

Discussion

The SCR is designed to detect children with SDB and AHI > 1 episode/h. It effectively excludes children without OSAS where treatment is not required. It is essential to diagnose children with moderate or severe OSAS even if there are not enough resources to perform PSG. This group of patients is at risk of persistent disease and benefits the most from treatment [2]. In this study, we determined the value of the SCR in identifying subjects with moderate-to-severe OSAS (AHI > 5). We have found that the threshold value of 6.25 has a high NPV (100%) but a low PPV (30%). We then calculated the cutoff value by which the questionnaire would have the optimal PPV and NPV for the diagnosis of subjects with moderate-to-severe OSAS (AHI > 5 episodes/h). The value of 8.25 gives the best combination of PPV (36%) and NPV (95%). In cases where a more reliable diagnostic method such as PSG is unavailable, SCR can be used to exclude moderate-to-severe OSAS diagnosis accurately.

Over the last 30 years, several screening questionnaires for pediatric OSAS have been published. Brouilette et al. [5] have described an OSA (obstructive sleep apnea) questionnaire assessing the observed apneas, difficulty breathing, and snoring. An OSA score ≥ 3.5 is considered diagnostic of OSA requiring adenotonsillectomy, and a score ≤ − 1

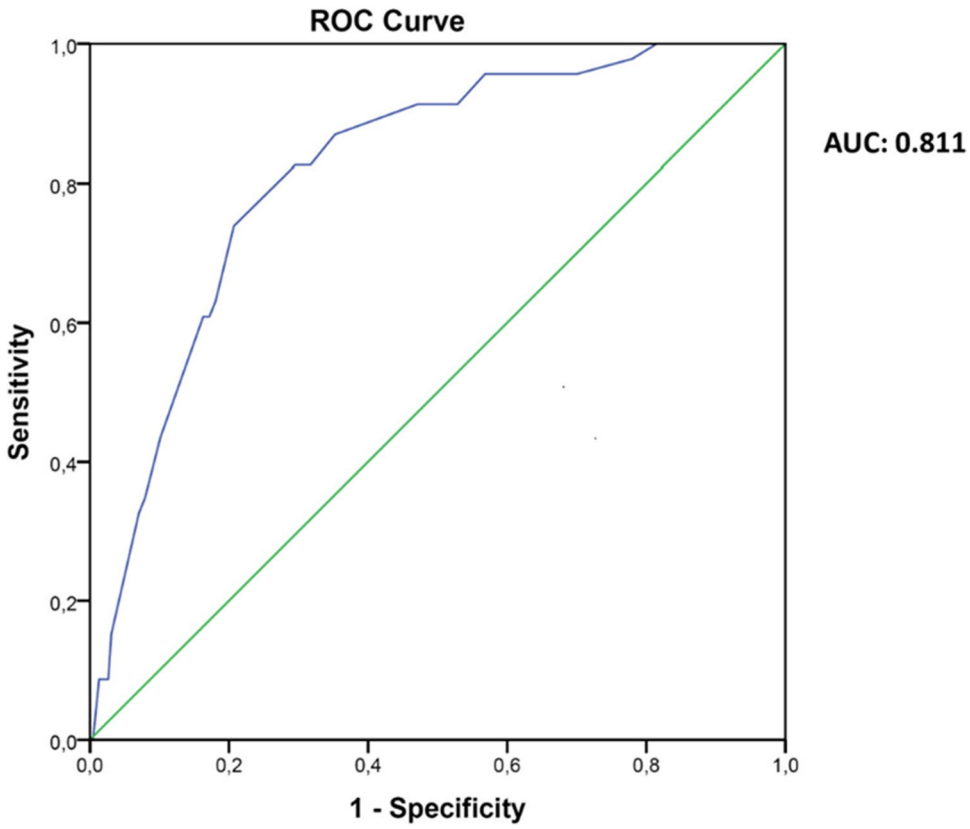

Fig. 1 ROC curve analysis of SCR score in predicting AHI > 5 episodes/h. AUC and its corresponding 95% confidence interval was 0.811 (0.747–0.876) with $P < 0.001$. ROC, receiver operating characteristic; SCR, sleep clinical record; AHI, apnea–hypopnea index; AUC, area under curve

is supposed to rule out sleep apnea, whereas a score between − 1 and 3.5 is considered uncertain. Three studies have been conducted to investigate the diagnostic accuracy of the OSA score. Carroll et al. [14] have concluded that the OSA score could not accurately distinguish between children with PS and subjects with OSAS. Rosen demonstrated that OSA score ≥ 3.5 had a poor sensitivity of 47%, a specificity of 28%, a PPV of 76%, and a NPV of 67% for OSAS diagnosis [15]. Half of the subjects (47%) had an uncertain symptom score. According to Lamm et al., the sensitivity of the OSA score was just 46%, but the specificity of 83% was excellent for predicting OSAS. Most of the patients (61%) had indeterminate values [16].

The pediatric sleep questionnaire (PSQ) includes 22 items [6]. It consists of three symptom categories: snoring, daytime sleepiness, and hyperactive behavior. A cutoff value of 0.33 is utilized to diagnose OSAS based on AHI ≥ 1 episode/h. Five studies assessed the diagnostic accuracy of the PSQ [17–21]. The PSQ questionnaire's sensitivity ranged from 50 to 84% and specificity ranged from 13 to 72%. Based on the results of one study [18], the sensitivity of the PSQ to diagnose subjects with moderate or severe OSAS is high (88%), but the specificity is low (17%).

The Severity Score is a six-item questionnaire [22]. A cutoff score of 2.72 might be used to differentiate children with AHI ≥ 3 from those with AHI < 3. The questionnaire had a sensitivity of 59%, a specificity of 83%, a PPV of 35%, and an NPV of 93%. Three studies evaluated the diagnostic accuracy of the Severity Score. Two of them mentioned that the Severity Score questionnaire had low sensitivities of 20% and 23%, respectively, for predicting children with AHI > 1.5 episodes/h, using a cutoff score of 2.72 [23, 24]. When the authors used a cutoff score ≥ 1, they found that diagnosing moderate and severe OSAS (AHI > 5 episodes/h) had sensitivities of 83% and 82%, respectively, and specificities of 64% and 50%. However, Nguyen et al., using a cutoff score of 2.75, demonstrated that the Severity Score showed high sensitivity (82%) and specificity (81%) for detecting moderate-to-severe OSAS [25].

OSA-18 questionnaire has been devised to determine the quality of children's lives with OSAS [8]. Four studies [26–29] have evaluated the accuracy of the OSA-18 questionnaire. The sensitivity of this tool to diagnose subjects with OSAS (AHI ≥ 1 or 2 episodes/h) has ranged from 55 to 63% and specificity between 40 and 84%. When used to diagnose children with AHI ≥ 5 episodes/h, the questionnaire had a sensitivity of 56 to 94% and 25 to 55% specificity. Walter et al. [29] have found that OSA-18 is extremely sensitive (93%) but has low specificity (25%) for the existence of moderate-to-severe OSAS.

Two more questionnaires have been evaluated for their ability to identify children with moderate or severe apnea. Chan et al. [30] have evaluated a modified Edward Sleepiness Scale and found that an excessive daytime sleepiness score > 8 could predict moderate or severe sleep apnea with a very low sensitivity of 29% and a high specificity of 91%. Wu et al. [31] have developed a new diagnostic scale that includes three items: snoring/gasping history, tonsillar size, and adenoidal to nasopharyngeal ratio. The diagnostic scale has high sensitivity (96%) but poor specificity (18%).

Of all the above questionnaires, in which their ability to predict the existence of moderate-to-severe OSAS in children has been assessed, only the Severity Score, in one of the three studies that evaluated it, proved useful (sensitivity + specificity at least 1.5) [32]. Our study assessed the accuracy of the SCR and compared it to objective PSG data. SCR with a cutoff value > 8.25 proved useful (sensitivity + specificity 1.53) to identify subjects with moderate-to-severe OSAS (AHI ≥ 5 episodes/h).

Villa et al. have proposed the SCR in 2013 [7]. Initially, a group of 279 children was studied. It was found to have an AUC of 0.80 (95% CI: 0.74–0.87), a high sensitivity (96%), and a moderate specificity (67%) for detecting children with AHI ≥ 1. The PPV was 88% and NPV was 86%. The SCR's ability to diagnose subjects with AHI ≥ 1 episodes/h has been tested in a later study [9] and it has been proven to have excellent sensitivity (92%) but low specificity (41%). The PPV was 92% and NPV was 41%. Subsequently, the researchers have used overnight pulse oximetry with McGill oximetry score > 1 in conjunction with the SCR ≥ 6.5 to identify children with AHI ≥ 5. The combined use of nocturnal pulse oximetry and SCR provides low sensitivity (39%), high specificity (97%), great PPV (94%), and fair NPV (61%). In our study, we showed that SCR alone with a cutoff value > 8.25 has much better sensitivity (83%) and NPV (95%) than the combined use of SCR ≥ 6.5 and nocturnal pulse oximetry. The specificity (71%) remains acceptable.

There are several limitations to this study that should be noted. The fact that our study was performed on a group of children who were referred to our sleep laboratory is a major potential limitation. The majority of subjects (169/273, or 61.9%) had an AHI ≥ 1 episode/h, indicating that they had pediatric OSAS. Because of the prevalence of OSAS, both positive and negative predictive assessments are likely to be affected. Another limitation is the lack of additional OSAS-related outcome measures, such as whether the SCR scale can predict OSAS complications. The strengths of this study are the large patient sample it includes, the prospective design, and the full-night PSG measurement.

In conclusion, the SCR is not intended to substitute PSG for diagnosing OSAS in children. The sensitivity of SCR with a cutoff value of 8.25 is high and the specificity is acceptable for recognizing moderate-to-severe OSAS. These results suggest that the SCR may be a useful and acceptable solution to identify subjects with AHI ≥ 5 episodes/h. However, further studies are necessary to determine whether

the relative efficacy of treatment differs depending on the diagnostic method and whether SCR is a valuable monitoring tool.

Declarations

Ethical approval The Larissa University Hospital Scientific Council gave its approval to this research (6/13–03-2018). This study was carried out in line with the 1964 Declaration of Helsinki and its later revisions, or similar ethical norms.

Informed consent All parents or legal guardians provided written informed consent.

Conflict of interest The authors declare no competing interests.

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