Original Article

Diagnostic Properties Modified OSA-18 Questionnaire in Children with Severe Obstructive Sleep Apnea

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Objective: To determine the diagnostic properties of OSA-18 and its modified version for detection of severe obstructive sleep apnea [OSA] in snoring children.

Materials and Methods: The present cross-sectional study was conducted in 123 patients (82 boys and 41 girls), aged younger than 12 years, who had snoring problems and performed polysomnography [PSG] at Siriraj Hospital. Those with incomplete questionnaires and inadequate PSG data were excluded. The patients were divided into two groups, non-severe OSA (apnea-hypopnea index [AHI] lower than 10) and severe OSA (AHI of 10 and above). The scores of OSA-18 questionnaires were compared between the two groups. Five most important questions (one from each domain) were selected to develop a modified shorter version of OSA-18.

Results: There was no statistically significant difference in total scores of OSA-18 between severe OSA and non-severe OSA groups. The optimal cut-off score (65) was selected from receiver operating characteristic [ROC]. The original OSA-18 had the specificity of 76%, the sensitivity of 40%, positive predictive value [PPV] of 40%, negative predictive value [NPV] of 76%, accuracy of 66%, and area under the curve [AUC] of 0.59. Meanwhile, at the optimal cut-off score (21), the modified OSA-18 had the specificity of 92%, the sensitivity of 34.3%, PPV of 63.2%, NPV of 77.9%, the accuracy of 76%, and AUC of 0.67, which was better than its original.

Conclusion: The modified shorter version of OSA-18 questionnaire with the optimal cut-off score of 21 is more useful, based on its high specificity, to enable physicians to quickly identify patients who require urgent treatment. Nevertheless, further studies of this newer version in different populations is recommended.

Keywords: OSA-18, Children, Screening, Severe obstructive sleep apnea

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Obstructive sleep apnea [OSA] is a common disorder involving partial blockage or complete obstruction of airflow in the upper airway during sleep, which leads to hypoxemia, hypercapnia, and poor sleep quality[1]. Previous epidemiologic studies showed that 5% to 12% of children had habitual snoring and 1.2% to 5.7% had OSA[2]. In Thailand, Anuntaseree et al reported the prevalence rates of habitual snoring and OSA in children of 6.9% to 8.5% and 0.7% to 1.3%, respectively[3]. If left untreated or delayed, OSA, especially if severe, may lead to several complications in neurobehavioral, cardiovascular, endocrine, and metabolic systems[4].

Generally, OSA diagnosis in children[1,2,5] is carried out by history taking, physical examination with additional investigations including polysomnography [PSG], nocturnal oximetry[6,7], nocturnal video recording[5], and lateral skull radiograph[5]. Although PSG is considered the gold standard for diagnosis, several limitations such as its scarcity, high cost, long waiting list, and inconvenience have made it impractical for a substantial number of patients. Thus, simpler, cheaper, and reliable alternative methods such as questionnaires[1,5,8] are increasingly useful to provide timely diagnosis and appropriate treatment of OSA, aiming to prevent complications and improve the quality of life [QOL] of both patients and their family[1,4,9].

Although various questionnaires have been introduced to screen for OSA[9-14], their clinical usefulness is unclear and conflicting. None is yet considered the most effective[1]. The OSA-18, a set of questionnaires for QOL evaluation in children with OSA[9,12,14,15], has been considered a potentially useful screening tool for OSA diagnosis with the questions directly point towards patient's signs, symptoms, and caregiver concerns regarding OSA consequences. The previous study of Franco et al[9] in 61 children

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showed the relationship between the OSA-18 scores and respiratory disturbance index [RDI] was significant when potentially confounding factors were adjusted, especially in the sections of sleeping disturbance and caregiver concern. Another study of Kang et al[12] in 109 children confirmed the significant correlation between the OSA-18 scores and PSG parameters, especially in the domain of sleep disturbance, daytime function, caregiver concerns, and total scores. Its usefulness in determining factors most affecting the QOL of children with OSA was mentioned[15]. On the other hand, the study of Constantin et al[14] in 163 children revealed that OSA-18 was not a valid screening test due to its lower sensitivity (40%) than that of oxygen level in blood during sleep; however, the gold standard, PSG, was not used in this study. Given these few and conflicting results, the usefulness of OSA-18 questionnaire for screening children with OSA, is still inconclusive. There is also a need for a short screening questionnaire of OSA with good reliability and validity. For example, the studies of Lachanas et al[16], and Spruyt and Gozal[17] used the OSD-6 questionnaire comprising six questions to screen for patients at high-risk for sleep disordered breathing [SDB]. Hence, the present study aimed to evaluate the diagnostic properties of the OSA-18 in screening for children with severe OSA and to modify the original questionnaire into a newer and shorter version so that it could be applied better in clinical practice.

Materials and Methods

The present cross-sectional study was approved by the Siriraj Institutional Review Board [SIRB], Faculty of Medicine Siriraj Hospital, Mahidol University, Thailand. Prospective data collection was done between March 2014 and December 2015, with the informed consent secured from the participants' parents.

Subjects

After obtaining the history and physical examination, patients were enrolled in the present study. Inclusion criteria were the parents of snoring patients, aged less than 12 years, with PSG at Department of Otolaryngology, Siriraj Hospital. Exclusion criteria were those with incomplete response of the OSA-18 questionnaires, inadequate PSG data including total sleep time 2 hours or less or severe recording artifacts. The recruited patients were classified into two groups based on the apnea-hypopnea index [AHI], i.e., non-severe OSA (AHI of less than 10) and severe OSA (AHI of 10 or more).

OSA-18 questionnaire and its modified version

The original version of OSA-18 questionnaire comprised 18 items distributed in five different sections including sleep disturbances (four items), physical abnormalities (four items), mental abnormalities (three items), daytime functions (three items), and caregiver concern (four items). With the range of score in each item from 1 to 7, the total scores would be from 18 to 126. Originally developed for QOL evaluation in OSA patients, the score less than 60 suggested a small impact on health-related QOL, whereas, the scores of 60 to 80 and a score of more than 80 defined a moderate impact and a large impact, respectively. In the present study, the authors used the validated Thai version of OSA-18 with permission[18]. The modified shorter version of the OSA-18 questionnaire was developed by selecting the single most defined item in five different sections after the original version was analyzed. The total score of our version were from 7 to 35. These five items showed the most significant difference in the mean scores between the severe and the non-severe OSA groups.

Polysomnography

All patients underwent an overnight, technician-attended, level I diagnostic PSG, (Compumedics, Somte, Profusion III; Victoria, Australia) at Siriraj Hospital. Apnea and hypopnea were defined according to the standard criteria recommended by the American Academy of Sleep Medicine version 2012. The technologists and sleep specialists were blinded to the patients' OSA-18 scores.

Outcome measurement

The diagnostic properties of the original and the modified versions of the OSA-18 questionnaire for detection of severe OSA included their sensitivity, specificity, positive predictive value [PPV], negative predictive value [NPV], accuracy, and area under the receiver operating characteristic [AUROC] curve. From these, we determined the optimal cut-off point for the diagnosis of severe OSA.

Statistical analysis

Continuous data were presented as mean ± SD and categorical data were presented as frequencies and percentages (%). Independent t-test or Mann-Whitney U test was used to compare continuous data, whereas Chi-square test was used to compare categorical data. Using PSG as the gold standard, the diagnostic properties of the original and modified versions of the OSA-18 for severe OSA were described in terms

of sensitivity, specificity, PPV, and NPV; results were presented with 95% confidence interval [CI]. To compare the diagnostic properties of the different screening models, an AUROC curve was calculated. The Predictive Analytics Software Statistics version 18.0 (New York, USA) was used for statistical analysis. The significance level was set at *p*-value of less than 0.05 in two-tailed tests.

Results

One hundred twenty-eight patients underwent the PSG between March 2014 and December 2015 were initially recruited. Five patients were excluded due to incomplete response of the questionnaires (n = 2), no rapid eye movement [REM] sleep in PSG (n = 2), and insufficient total sleep time (n = 1). Thus, the data from 123 patients (82 boys and 41 girls) with mean age of 7.0 years (range, 2 to 12 years) and mean AHI of 8.9 events/hour were analyzed. The averages of total sleep time and sleep efficiencies were 419.88 minutes and 90%, respectively, indicating good quality of sleep. According to the AHI, the patients were classified as 35 patients (28.5%) in severe OSA (AHI of 10 or more) group, and 88 patients (71.5%) in non-severe OSA group with seven patients (5.7%) (AHI of less than 1). The cut-off point at AHI of 1 or greater was selected to identify patients who required more urgent diagnosis (first priority for PSG or sleep studies) and treatment such as tonsillectomy. Further detailed data of the patients were shown in Table 1.

Regarding the scores of the OSA-18 questionnaire, the fifth section (caregiver concern) revealed the

Table 1. Data of patients according to non-severe and severe OSA

Data	Non-severe OSA[a] (n = 88)	Severe OSA[b] (n = 35)	*p*-value
Age (year)	7.0±2.4	6.8±2.7	0.804
Total sleep time (minute)	424.4±58.5	408.3±49.4	0.155
Sleep efficiency (%)	91.3±7.2	89.1±7.2	0.140
REM latency (minute)	120.0±55.5	119.2±66.9	0.949
Stage N1 (%)	5 (0.8 to 78.0)	8 (3.5 to 21.0)	0.050*
Stage N2 (%)	38.0±8.8	35.2±11.4	0.141
Stage N3 (%)	35.3±10.2	35.1±12.9	0.923
Stage R (%)	19.2±5.6	19.5±6.4	0.803
AHI (events/hour)	4.7±2.6	19.3±10.7	<0.001*
RDI (events/hour)	12.3±6.5	25.6±12.5	<0.001*
Apnea index	0.3 (0.0 to 3.0)	2.28 (0.0 to 30.7)	<0.001*
Minimal O_2 saturation	88.7±7.7	83.6±9.9	<0.007*
Time (%) O_2 >90%	97.5±10.8	96.1±13.8	0.577
Arousal index	11.4±6.3	19.4±5.6	<0.001*
Total questionnaire score	53.5±16.8	59.0±17.0	0.107

OSA = obstructive sleep apnea; REM = rapid eye movement; AHI = apnea-hypopnea index; RDI = respiratory effort-related arousals; N = non-REM; R = REM

[a] Non-severe OSA (AHI <10), [b] Severe OSA (AHI ≥10)

* Statistically significant (p<0.05)

Data presented as mean ± standard deviation or median (min-max)

Table 2. Comparison between OSA-18 score and AHI

OSA-18 questions	Non-severe OSA	Severe OSA	*p*-value
1. Disturbances during sleep			
1.1 Loud snoring	3.8±1.6	4.4±1.5	0.066
1.2 Intermittent breathing pauses	2.4±1.2	3.0±1.3	0.044*
1.3 Gasping or choking during sleep	2.5±1.2	2.8±1.3	0.207
1.4 Restlessness during sleep, wakes up suddenly	2.6±1.3	3.0±1.4	0.162
Total score of section 1	11.4±4.4	13.3±4.7	0.047*
2. Physical abnormalities			
2.1 Breathing through the mouth due to inconvenient nose breathing	3.1±1.5	4.2±1.7	0.002*
2.2 Frequent colds or upper respiratory tract infection	3.3±1.4	3.4±1.4	0.537
2.3 Runny nose	3.0±1.2	3.2±1.2	0.446
2.4 Difficulty swallowing	1.9±1.3	1.8±0.8	0.666
Total score of section 2	11.5±4.4	12.8±4.0	0.140
3. Mental abnormalities			
3.1 Mood swings or screaming	2.0±1.3	2.6±1.4	0.016*
3.2 Aggressive and disruptive behavior	2.4±1.6	2.5±1.3	0.790
3.3 Discipline problems, difficult to manage behavior	2.4±1.3	2.8±1.2	0.189
Total score of section 3	6.9±3.8	8.0±3.6	0.787
4. Effects on daytime function			
4.1 Daytime sleepiness or fatigue	2.0±1.0	2.3±1.2	0.306
4.2 Trouble concentrating	2.8±1.5	2.7±1.5	0.650
4.3 Trouble getting up in the morning	3.1±1.8	2.8±1.7	0.453
Total score of section 4	8.0±3.3	7.8±3.7	0.785
5. Caregiver concern			
5.1 Concern about the child's general health	4.9±1.6	5.1±1.4	0.509
5.2 Concern about the child's insufficient breathing	4.6±1.7	5.4±1.3	0.028*
5.3 Impact on work and other daily routine	2.6±1.4	3.0±1.5	0.238
5.4 Feeling upset and anxious about the problem	3.2±1.5	3.3±1.6	0.675
Total score of section 5	15.5±5.4	16.9±4.9	0.178
Total score	53.5±16.8	59.0±17.0	0.107

OSA = obstructive sleep apnea; AHI = apnea-hypopnea index

* Statistically significant (p<0.05)

Data presented as mean ± standard deviation

Table 3. Questionnaire of modified short version OSA-18

Scores	1	2	3	4	5	6	7
Items	None	Barely	Sometimes	Quite often	Often	Usually	Always
1. Intermittent breathing pauses							
2. Breathing through the mouth due to inconvenient nose breathing							
3. Mood swings or screaming							
4. Daytime sleepiness or fatigue							
5. Concern about the child's insufficient breathing							
Total score							

OSA = obstructive sleep apnea

Table 4. Diagnostic properties of OSA-18 and modified short version OSA-18

Properties	OSA-18 (95% CI)	Modified short version OSA-18 (95% CI)
Specificity	0.76 (0.66 to 0.84)	0.92 (0.84 to 0.96)
Sensitivity	0.40 (0.26 to 0.56)	0.34 (0.21 to 0.51)
PPV	0.40 (0.26 to 0.56)	0.63 (0.41 to 0.81)
NPV	0.76 (0.66 to 0.84)	0.78 (0.69 to 0.85)
Accuracy	0.66 (0.57 to 0.74)	0.76 (0.64 to 0.82)
AUC	0.59 (0.47 to 0.70)	0.67 (0.56 to 0.78)

OSA = obstructive sleep apnea; PPV = positive predictive value; NPV = negative predictive value; AUC = area under the curve

highest mean total score (15.93) whereas, the third section (mental abnormalities) showed the lowest mean total score (7.26). Comparison between the severe OSA and the non-severe OSA groups indicated that all sections, except the fourth section (effects on daytime function), had at least one question with statistical significance between the two groups (Table 2). These distinct questions and the item of daytime sleepiness or fatigue in the fourth section were chosen to develop a shorter version for more convenience and higher specificity. These items of the modified OSA-18 questionnaire are presented in Table 3. Based on the total scores from the shorter version, there was a significant difference between the severe and the non-severe OSA groups (p<0.001). The diagnostic properties of the original and the modified OSA-18 were compared including sensitivity, specificity, PPV, NPV, and accuracy (Table 4). The best cut-off score of 65 and 21 of the original and the modified versions were identified from their ROC curve (Figure 1, 2).

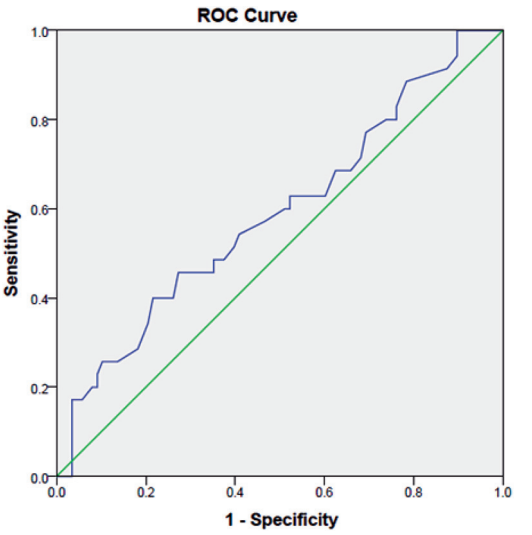

Figure 1. ROC analysis of the total score obtained from OSA-18.

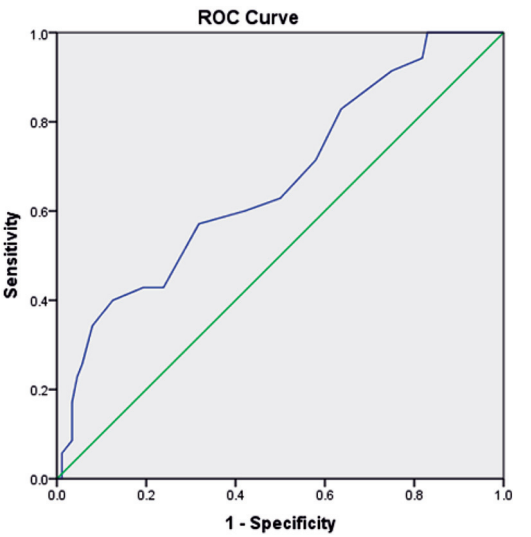

Figure 2. ROC analysis of the total score obtained from the modified short version OSA-18.

Discussion

Without proper treatment, severe OSA can lead to several complications. Although PSG is the accepted gold standard of the diagnostic test, there are still limitations such as its scarce availability due to the long

waiting list and high cost. Therefore, simpler, cheaper, and faster screening tools are increasingly needed for children, especially if suspected for severe OSA, which requires urgent diagnosis and treatment. In our study, there was no statistical significant difference in the total score of the original OSA-18 questionnaire between the severe OSA and the non-severe OSA groups. Nonetheless, the scores in some sections, particularly on sleeping disturbances demonstrated the greatest difference in both groups of patients, consistent with those of Franco et al[9] and Kang et al[12]. The items with most significant differences between the severe and the non-severe OSA groups included intermittent breathing pauses, breathing through the mouth due to inconvenient nose breathing, mood swings or screaming, and concern about the child's insufficient breathing. Even though daytime sleepiness or fatigue showed no statistical significant difference, but it can be the most common and important symptoms in OSA patients that affects their QOL. However, this item should be included like those previously mentioned items to develop and modify into our shorter version of the OSA-18 questionnaire.

Following the diagnostic properties in our series, the original OSA-18 had the specificity of only 76%, and sensitivity of 40% when using the optimal cut-off score of 65 from ROC curve. These results were consistent with those of Borgström et al[19] and Ishman et al[20] stating the low accuracy of the original OSA-18 questionnaire for OSA patient screening. Nonetheless, the modified shorter version of OSA-18 had its specificity as high as 92%, but with the sensitivity of only 34.3% when using the optimal cut-off score of 21 from ROC curve. With this high specificity, our shorter version could precisely confirm that the snoring children with high score had a high-risk for severe OSA, which required urgent treatment. Furthermore, our short questionnaire could be more convenient for patients' caregivers and physicians to complete and interpret. Moreover, due to the poor sensitivity of our questionnaire, it should not be used for OSA screening in general children population, particularly if they have no sleep complaints.

In the meantime, there were some limitations in the present study. First, the analysis of the diagnostic properties of our newly modified shorter version of the OSA-18 questionnaire was performed in the same groups of OSA patients who previously completed the OSA-18 questionnaire. Further studies of this newer version in various populations should be done. Secondly, our study was aimed to screen the children with severe OSA that urgently seek medical consultation in special clinics. Thus, our results may not be suitably applied for the screening of OSA in general population.

Conclusion

With the acceptable specificity, our newly modified shorter version of OSA-18 questionnaire is useful for quick identification of those requiring urgent treatment. It also enable the differentiation of the severe OSA patients from the non-severe OSA. Further studies should be done using different group of children to confirm its validity in the general population.

What is already known on this topic?

The OSA-18 questionnaire for screening of children with severe OSA.

What this study adds?

Success rate of the modified shorter versions of the OSA-18 questionnaire for screening of children with severe OSA.

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Potential conflicts of interest

The authors declare no conflict of interest.

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