Validity and Reliability of the Thai Version of the Pediatric Obstructive Sleep Apnea Screening Tool

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Abstract

Objective: Obstructive sleep apnea (OSA) is highly prevalent in children and requires an expensive and relatively unavailable sleep study for diagnosis. This study was undertaken to translate the previously validated OSA screening tool (POSAST) to the Thai language and assess its accuracy and test-retest reliability in at-risk symptomatic children.

Study design: Prospective cross-sectional cohort study

Methods: Pediatric patients clinically referred for suspected OSA who underwent overnight polysomnography *(*PSG*)* were recruited, and caregivers completed the Thai version of the POSAST. The same questionnaire was completed again after 2-4 weeks.

Results: One hundred and ten subjects completed the study. The mean age was 8.4±2.9 years. The mean apnea-hypopnea index *(*AHI*)* was 10.9±11.9 events/hour. Test-retest reliability *(*Pearson correlation coefficient = 0.96, $P<0.001$*)* and internal consistency *(*Cronbach's alpha coefficient = 0.82, $P<0.001$*)* between each question were excellent. A cumulative equation-derived score cut-off of 1.9 yielded 78.4% sensitivity and 50.0% specificity, while a numerical additive score cut-off of 8 corresponded to 81.1% sensitivity and 52.8% specificity for diagnosing moderate and severe OSA *(*AHI ≥5 events per hour*)*

Conclusion*:* The internal consistency and reproducibility of the Thai version of the POSAST are satisfactory, display acceptable validity, and the instrument can be used for screening symptomatic Thai children for OSA.

Keywords: obstructive sleep apnea, pediatric, questionnaire, Thailand

Introduction

Obstructive sleep apnea (OSA) is characterized by intermittent partial or complete obstruction of the upper respiratory tract during sleep, which may induce the emergence of hypoxia and hypercapnia, as well as repetitive episodes of arousal to reestablish the airway patency.[1] The prevalence of OSA has been estimated around 1.2% to 5.7% of the pediatric population in the United States[2] and 0.7% to 1.3% among the children of Thailand.[3] However, the frequency of symptoms suggestive of OSA is much higher, and can reach 10-27%, depending on the criteria used.[4-6] Adverse consequences of pediatric OSA include cardiovascular abnormalities and neurocognitive and behavioral dysfunction, with inevitably adversely affect the quality of life[7-10] Therefore, early diagnosis and treatment of pediatric OSA are essential to prevent these potentially lifelong adverse outcomes.

Overnight polysomnography (PSG) is considered the gold standard for the diagnosis of OSA.[11-14] However, it incurs high costs and long waiting times in many countries, and the need for appropriately sleep technologists and sleep medicine physicians limit PSG widespread accessibility, especially in Thailand, where resources are limited. According to the American Academy of Pediatrics Consensus guidelines (AAP), screening for snoring and other OSA-related symptoms is recommended in every child.[14]

In an effort to identify pediatric patients who are at risk of OSA, multiple studies have emerged, and have focused on the development of a questionnaire tool.[15-18] However, the majority of these questionnaires is generally too lengthy and time-consuming to be used as a screening tool by primary care physicians in their customary busy practices, since each of such questionnaires includes between 22 to 40 questions.[2, 19] In addition, most of these questionnaires do not accurately predict a diagnosis of pediatric OSA as collaborated by a PSG.[17]

As a potential solution, Spruyt and Gozal conducted a comprehensive machine learning analysis of a large number of *a priori* relevant questions related to pediatric OSA symptoms, and a set of 6 short, hierarchically arranged questions was proposed as a new screening instrument, the Pediatric Obstructive Apnea Screening Tool (POSAST). The POSAST brief and easy to use in clinical practice, [20] and was subsequently validated in the English language, [18] whereby using a cumulative score ≥1 yielded a sensitivity of 83%, a specificity of 64%, PPV of 28%, and NPV of 96% for diagnosing moderate and severe OSA (apnea-hypopnea index, AHI ≥5 events/hr). The POSAST was also translated and validated into Japanese and French languages.[21, 22] In the French version, a score of >2.72 exhibited an 82% sensitivity, 81% specificity, and 92% negative predictive value for detecting an AHI of ≥5 events/hr.[22]

In Thailand, the Thai Guideline for Childhood OSA was published by the Thai Sleep Society in 2013.[23] This guideline also emphasizes the critical importance for clinicians to screen for snoring and other OSA-related symptoms in every child. In the guideline, screening with the POSAST was also recommended. However, this questionnaire was not officially translated and validated in Thailand. Therefore, the objectives of this study were to translate the POSAST to the Thai language and to assess the validity and test-retest reliability of the Thai version of the questionnaire in symptomatic children.

Materials and Methods

Translation of Pediatric Obstructive Apnea Screening Tool to the Thai Language

After permission for translation was obtained, the actual translation of the questionnaire from English to the Thai language was conducted by two Thai native speakers with a good academic background in English. After comparing the two translations, incongruences were identified and

resolved by consensus. Backward translation from Thai to the English language was then performed by two English native speakers with fluency in Thai. The forward and backward translations were assessed by two pediatric otolaryngologists, and then a consensus was achieved to reach the final translation. Ten volunteers in the community were then recruited to ascertain that the questionnaire could be easily understood and that the content corresponded to the actual intent of the original text. The translated Thai version is shown in the Online Supplement.

The six questions were in the following order: (1) Do you shake your child to breathe? (2) Have you witnessed apnea during sleep? (3) Does your child struggle with breathing when asleep? (4) Do you have concerns about your child's breathing while asleep? (5) How loud does your child snore? and (6) Does your child snore while asleep? All questions were answered on a Likert-type response scale: never (0), rarely (once per week; 1), occasionally (twice per week; 2), frequently (three to four times per week; 3), and almost always (over four times per week; 4). The exception was question number five, where response options were: mildly quiet (0), medium loud (1), loud (2), very loud (3), and extremely loud (4) for the preceding 6-month time frame. The cumulative score of the questionnaire was calculated according to the formula (where Q1 = response to question 1, Q2 = response to question 2, and so on): $A = (Q1 + Q2)/2$; $B = (A + Q3)/2$; $C = (B + Q4)/2$; $D = (C + Q5)/2$; the cumulative score $= (D + Q6)/2$.[18]

However, to increase the convenience for physicians and medical personnel when using this questionnaire, we also calculated a total additive score (by using the formula: Q1+Q2+Q3+Q4+Q5+Q6). We investigated the diagnostic performances of the POSAST when using both the equation-derived score and the cumulative additive score.

Study design

This prospective study was conducted in the Department of Otorhinolaryngology, Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok, Thailand, from October 2014 to December 2016. All participants were consecutively recruited and scheduled for PSG evaluation according to the indications outlined in the Thai clinical practice guideline.[14, 23] Written informed consent was obtained from all caregivers. The protocol for this study was approved by the Siriraj Institutional Review Board (SIRB) (protocol#: Si 638/2014).

Cohort

Pediatric subjects aged 2 to 15 years with clinically suspected OSA (documented history of labored snoring, witnessed apnea, struggling, and/or gasping during sleep) who were referred to the Department of Otorhinolaryngology, Faculty of Medicine Siriraj Hospital and scheduled for an overnight PSG evaluation were included. Patients with comorbidities including craniofacial anomalies, neurological diseases, and cardiovascular diseases were excluded. All caregivers who answered the questionnaire could read and write Thai.

Intervention

Caregivers were asked to complete the Thai version of POSAST on the evening of the PSG test. All children underwent overnight level 1 PSG. A digital commercially available PSG system (Somte´ PSG, Compumedics Ltd, Victoria, Australia) was used to record sleep parameters, including electroencephalogram (EEG), electrooculogram (EOG), chin and leg electromyogram (EMG), electrocardiogram (ECG), airflow signals, respiratory effort signals, oxygen saturation, and body position. PSG recordings were scored and interpreted by sleep specialists using criteria set forth by

the American Association of Sleep Medicine (AASM).[24] Based on the apnea and hypopnea index (AHI), the PSG findings were classified for each child as normal (AHI<1 event/hr), mild OSA (1≤AHI<5 events/hr), moderate OSA (5≤AHI<10 events/hr), and severe OSA (AHI≥10 events/hr).[5] Sleep specialists were blinded to the POSAST results. The same questionnaire was completed again by the same caregivers in the next 2-4 weeks for test-retest reliability assessments.

Statistical analysis

Categorical data are presented as counts and percentages. Continuous data are shown as mean ± standard deviation (SD) for parametric variables and median and interquartile range (IQR) for nonparametric variables.

Comparison of categorical data between groups was done by using Chi-square or Fischer's exact tests. Comparison of continuous data between groups was conducted by using unpaired t-test for parametric variables and Mann-Whitney U test for nonparametric variables.

The reliability of the questionnaire was determined by internal consistency and test-retest reliability. Internal consistency for each of the questions was determined by calculating the Cronbach's alpha coefficient, categorized as follows: very satisfactory (0.80-0.89), good (0.50-0.79), lower than the original questionnaire < 0.5.[25] Test-retest reliability was evaluated by using the Pearson correlation coefficient (r), categorized as follows: little or none (0-0.24), fair (0.25-0.49), moderate to good (0.50-0.74), and very good to excellent (0.75-1.00).[26]

The validity of the questionnaire was assessed by constructing receiver operating characteristic (ROC) curves to identify the cumulative score and total additive score cutoff points (i.e., sensitivity, specificity, positive predictive value [PPV], negative predictive value [NPV], and

accuracy) that identify high risk for moderate and severe OSA (AHI of ≥5 events/hr).

Statistical significance was set as a two-tailed P-value of less than 0.05. All statistical analyses were performed using SPSS statistical software version 18 (SPSS, Inc IBM, Chicago, Illinois)

Results

We consecutively recruited 110 subjects aged 2 to 15 years with clinically suspected OSA who met the selection criteria. Their mean age was 8.4 ± 2.9 years, and their baseline demographic, nutritional status[27], and clinical characteristics are shown in **Table 1**. The mean AHI was 10.9±11.9 events/hr. Based on the PSG-derived AHI criteria, 36 children (32.7%) suffered from mild OSA, 37 (33.6) had moderate OSA, and 37 (33.6%) had severe OSA. No significant age differences emerged between mild OSA versus moderate and severe OSA cases (8.0±2.7 vs 8.5±3.0 years; *P*=0.403) The PSG measures in mild OSA and moderate and severe OSA groups are shown in **Table 2**, and as anticipated revealed significant differences in terms of AHI, nadir oxyhemoglobin saturation, and arousal index between the mild OSA group and the moderate and severe OSA group.

Since there were no children without OSA (AHI <1/hr) in this study, we used the AHI cutoff of 5 events/hr, to screen for moderate to severe disease. The ROC curve is shown in **Figure 1,** and displayed an area under the curve (AUC) of 0.627 for the equation-derived score (95th percentile confidence intervals, 95% CI: 0.512-0.743; *P*=0.031) and an AUC for the total additive score of 0.647 (95% CI: 0.530-0.764; *P*=0.013), respectively. The equation derived score cut-off set at 1.9 provided 78.4% sensitivity and 50.0% specificity, while an additive total score cut-off of 8 yielded 81.1% sensitivity and 52.8% specificity for identifying moderate and severe OSA *(***Table 3***)***.**

There was a very satisfactory internal consistency among each question of the questionnaire *(*Cronbach's alpha coefficient = 0.82, *P*<0.001*)*. In terms of test-retest reliability, the mean time elapsed between completion of the questionnaire by the caregivers was 20±7 days. There was an excellent test-retest reliability *(*Pearson correlation coefficient, r = 0.96, *P*<0.001*)*. The scores of each question were compared between mild OSA group versus moderate and severe OSA group ***(*Table 4*)***. There was a significant difference in four out of six questions between both groups. The diagnostic values are summarized in **Table 5** along with comparisons with previous studies.

Discussion

This study demonstrated the validity and reliability of the Thai version of POSAST in the process of screening snoring symptomatic children. The original version of POSAST was published in the English language in 2012, as a clinical severity scale consisting of a set of 6 hierarchically arranged questions for the screening of children such as to define those at high risk for OSA.[20] The cut-off score was set at 2.72 and was successful at discriminating community non-clinical referral children with AHI ≥3 from those with AHI < 3, as illustrated by an AUC 0.79±0.03 (95%CI 0.76-0.81), a sensitivity of 59.03% (95%CI 50.5%-67.1%), and a specificity of 82.85% (95%CI 80.2%-85.3%). The POSAST was further evaluated by Kadmon *et al* in a clinical referral cohort.[18] In this setting, the original cutoff score of 2.72 displayed sub-optimal sensitivity and specificity when targeting AHI values of >1 or > 1.5 events/hr. In contrast, when using the POSAST with a score set at 1 for diagnosing moderate and severe OSA (AHI ≥5 events/hr), the AUC obtained from the ROC curve was 0.647, with a sensitivity of 83%, a specificity of 64%, PPV of 28%, and NPV of 96%.[18] These differences in POSAST accuracy were attributed to the different types of cohort, i.e., low and high pre-test probability community and clinical referral samples, respectively. In other words, pediatric patients referred to the sleep clinic tend to have more symptoms suggestive of a sleep

disorder, such that even milder nocturnal breathing symptoms appear to be sufficient to reliably detect OSA. Hence, a lower cutoff threshold was needed.[18] The POSAST was also translated to Japanese and French languages.[21, 22] In the French version, the cumulative score was named as severity hierarchy score (SHS). An SHS score of > 2.75 exhibited an 82% sensitivity, 81% specificity, and 92% negative predictive value for detecting an AHI of ≥5 events/hr. The AUC obtained from the ROC curve was 0.87. Thus, the POSAST has consistently demonstrated its internal and external validity across different settings, prompting our interest in adapting it to the Thai population.

In this study, we included 110 subjects of similar age to previous studies, a sample size that exceeded the ones included in previous studies and deemed as sufficient to provide the robustness required for implementation in clinical settings.[18, 22]

The present study also used the AHI cut-off at 5 events/hr to differentiate moderate and severe OSA from mild OSA, since this AHI cut-off value has associated with substantial increases in the risk of cardiovascular and cognitive morbidities.[9, 28, 29] The AUC of the cumulative Thai POSAST score for diagnosing moderate and severe OSA was 0.627, i.e., comparable to the English versions of the instrument.[18, 22]

Since the severity of OSA was greater in our cohort *(*67.3% has AHI ≥5 events/hr*)*, the higher pre-test probability may have improved the screening accuracy of the tool in our setting [18, 22]. Similarly, the reduced proportion of either children who snore but have normal AHI values or those with mild disease could have reduced the discriminative ability of the instrument, an assumption that will require expanded testing in the near future.

Although the Thai version of the questionnaire has acceptable sensitivity for screening

patients in the clinic who requires more urgent management, either undergo PSG or surgical intervention, cultural and language differences may also impact the accuracy of the questionnaire. Accordingly, we also investigated whether the total score sum can be used instead of the original score since such an approach facilitates the ease of implementation. Since the AUC and other predictive metrics were essentially comparable using the two methods, the sum score can also be used for identifying children at high risk of OSA. Finally, we undertook the initiative to assess test-retest reliability and consistency, and the favorable findings further collaborate the potential use of the POSAST in primary care settings, whereby it can be filled out during the routine periodic well-child visits.

Some limitations of the study deserve comment. First, the patients in our cohort were more likely to suffer from more severe OSA in the previous studies, which may either reflect the fact that the study was conducted in a tertiary hospital, or may also reflect the fact that awareness of pediatric OSA is lower in Thailand, and therefore only more severe children are referred by their primary care physicians. Secondly, we did not evaluate the POSAST after treatment for OSA to ascertain changes corresponding to improvements in the severity of OSA or whether the instrument can readily detect residual OSA after adenotonsillectomy. Finally, future testing of this questionnaire in community settings or selected pediatric specialized clinics *(*e.g., children with ADHD, asthma, or Down syndrome*)* appears to be a worthwhile endeavor.

Conclusion

The Thai version of the POSAST demonstrates satisfactory internal consistency and reproducibility with acceptable validity. Both cumulative and total scores can be used for the prediction of patients with moderate and severe OSA. This set of questions is simple and easy to use

as a screening tool for OSA in Thai children.

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Table and Figure Legend

Table 1. Baseline Demographic and Clinical Characteristics **(**n = 110**).**

Table 2. Comparison of Sleep Parameters between Mild OSA Group (AHI < 5 events per hour) versus Moderate and Severe OSA Group **(**AHI ≥ 5 events per hour**).**

Table 3. Diagnostic Capability of the Pediatric Obstructive Sleep Apnea Screening Tool for Diagnosing Moderate and Severe OSA (AHI ≥ 5 events per hour).

Table 4. Comparison of the Score of Each Question between Mild OSA Group versus Moderate and Severe OSA Group.

Table 5. Cut-off Value of the Cumulative Score and Diagnostic Performance of Pediatric Obstructive Sleep Apnea Screening Tool for Moderate and Severe OSA (AHI ≥ 5 events per hour).

Figure 1. Receiver operating characteristic (ROC) curve for predicting moderate and severe obstructive sleep apnea (AHI≥ 5) according to the cumulative and total score. Area under

the curve (AUC) of cumulative score: 0.627 (95% confidence interval, 0.512-0.743; *P*=0.031). Area under the curve (AUC) of total score: 0.647 (95% confidence interval, 0.530-0.764; *P*=0.013).