Original article

The reliability and validity of the Thai version Epworth sleepiness scale for children and adolescents (ESS-CHAD) in pediatric obstructive sleep apnea

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Abstract:

Background: *Obstructive sleep apnea (OSA) is a sleep disorder that the upper airway collapses momentarily while sleeping, which leads to many symptoms, e.g. snoring, excessive daytime sleepiness, learning difficulties, and behavioral problems. On account of high global prevalence of OSA, many questionnaires were used to screen OSA patients. Epworth sleepiness scale (ESS) was successfully developed for screening excessive daytime sleepiness in adults. Subsequently, due to insufficient screening tools in children, ESS-CHAD was recently created.* ***Objective:*** *The objectives of this study are to evaluate the reliability and validity between Epworth sleepiness scale for children and adolescents (ESS-CHAD) and pediatric obstructive sleep apnea, and to investigate the correlation between Epworth sleepiness scale for children and adolescents (ESS-CHAD) and the severity of pediatric obstructive sleep apnea.* ***Methods:*** *A total of 41 subjects (31 males and 10 females) were recruited. In order to check the discriminant validity of the ESS-CHAD, we included 26 patients with obstructive sleep apnea (OSA) confirmed by polysomnography, 9 patients with mild OSA, 10 patients with moderate OSA, and 7 patients with severe OSA. The test-retest reliability was investigated in 41 subjects to check the responsiveness properties of the questionnaire.* ***Results:*** *The internal consistency demonstrated by Cronbach's alpha coefficients for standardized item was 0.836. The test-retest reliability was shown by intra-class correlation coefficients of 0.982. There was a statistically significant difference between the mean of the ESS-CHAD scores of the mild OSA (3.33±1.87) and the severe OSA patients (9.85±5.43) ($p < 0.05$). Also, there was a statistically significant difference between the mean of the ESS-CHAD scores of the Pediatric Sleep Questionnairre (PSQ) negative patients (4.31±2.72) and the PSQ positive patients (7.36±5.74) ($p < 0.05$). The ESS-CHAD score had the highest sensitivity of 70.59% and the highest specificity of 77.78% with a cut-off score ≥ 5. However, there was no statistically significant difference in the correlation between ESS-CHAD scores and polysomnogram parameters.* ***Conclusions****: Our first Thai version of the ESS-CHAD showed an excellent internal consistency and test–retest reliability. It is able to discriminate between mild OSA patients and severe OSA patients with a cut-off value ≥ 5; however, it does not have a correlation with the polysomnogram parameters. Therefore, we recommend use it to combine with more comprehensive clinical evaluation in pediatric obstructive sleep disordered breathing patients.*

Keywords: ● *Epworth sleepiness scale for children and adolescents* ● *Obstructive sleep disordered breathing* ● *Reliability and validity* ● *Thai version* ● *Pediatric obstructive sleep apnea*

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Introduction

Obstructive sleep apnea (OSA) is the condition when upper respiratory tract momentarily collapses during sleep, which leads to many symptoms, eg. snoring, excessive daytime sleepiness, learning difficulties, and behavioral problems[2,14]. Globally, the prevalence of habitual snoring is 2.4-17.16%, and the prevalence of OSA is 1.2-5.71%[1]. In Thai children, the prevalence of habitual snoring is 6.9-8.5%, and the prevalence of OSA is 0.7-1.3%[1]. Untreated OSA would affect patient's neurobehavioral system, cardiovascular system, endocrine and metabolic systems, and also the quality of the patient's life[2,16-18]. Although, the gold standard for diagnosis OSA is polysomnography (PSG)[24,25], there are many limitations to access the test such as many specific and limited equipment, requiring special staffs and expensive costs, which result in delaying test and diagnosis. There were many attempts to establish screening tools for screening OSA such as overnight pulse oximetry, nocturnal video recording, ambulatory PSG, and pediatric sleep questionnaires[19-25].

In adults patients who have excessive daytime sleepiness, there are many questionnaires developed for screening OSA[5-8]. Epworth sleepiness scale (ESS) questionnaires was developed by Murray W. Johns in 1991[3,4]. Many studies showed high reliability and validity for screening excessive daytime sleepiness in patients with narcolepsy, idiopathic hypersomnia, and OSA using this questionnaires[26]. The sensitivity was 97% and the specificity was 100%[4]. The ESS was translated into a Thai version questionnaire by Wish Banhiran[9], and the study revealed that the Thai version ESS questionnaire was able to discriminate between normal people and OSA patients.

In children and adolescents with OSA, they presented snoring, excessive daytime sleepiness (EDS), behavioral problems, and learning difficulties[27,28]. These symptoms were different from those in adults, hence, some questions in ESS questionnaire were not suitable for screening pediatric OSA. Subsequently, due to insufficient screening tools in children, Epworth sleepiness scale for children and adolescents (ESS-CHAD) was created. From the study in Australia, conducted by Y Grace Wang[11] and Kitty C Janssen[12], ESS-CHAD had high reliability and validity in identifying excessive daytime sleepiness conditions in children and adolescents, which was a part of pediatrics OSA clinical manifestations. Although ESS-CHAD has been used in several sleep researches and clinical practices for years, there have never been any study to validate the Thai version of the ESS-CHAD and its application in pediatric OSA patients. The objectives of this study are to translate the ESS-CHAD into Thai language, to evaluate the reliability and validity between the Thai version ESS-CHAD and pediatric obstructive sleep apnea, and to investigate the correlation between the Thai version ESS-CHAD and severity of pediatric obstructive sleep apnea.

Materials and Methods

This study was supported by the Research Management Fund and Sleep disorder center, Department of Pediatrics, Phramongkutklao Hospital, and was conducted between January 2021 and May 2022 after an approval from the Phramongkutklao Institutional Review Board, Royal Thai Army Medical Department. The translation processes were also kindly permitted by the Mapi Research Institute in Lyon (Dr. James W. Varni).

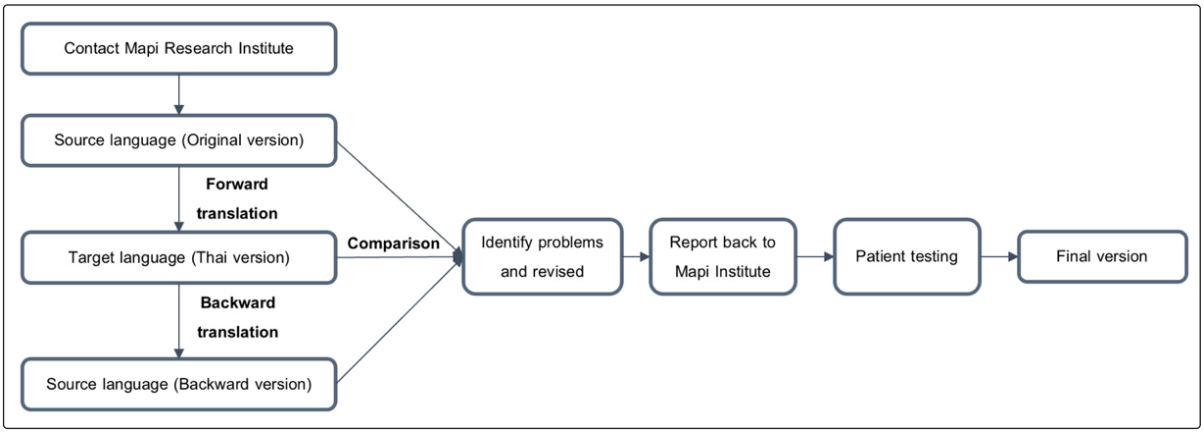

Figure 1 Flow diagram of the translation and validation process

Translation of the original ESS-CHAD into Thai language

The translation of the Australian English version of the ESS-CHAD (copyright of M.W. Johns 1990, 1997, 2013) which was used in this study followed standardized processes. These started from the translation of the ESS-CHAD English version into Thai by four translators who are fluent in English, including one professional translator from a university. One of these translated versions was blindly selected with total agreement by the research committees who are medical specialists and translated back into English by another professional translator for comparison. This process was repeated until the selected final English version is as close as possible in vocabulary and meaning to the original. The final version was then tested in a small group of subjects and minimally adjusted before applying it to the larger study groups.

Participants

A total of 41 subjects (31 males and 10 females) were recruited. The inclusion criteria were children with age between 7-17 years old who received treatment at Phramongkutklao Hospital with signs and symptoms that are suspected OSA and would be tested with Polysomnogram, and participants and their guardians intend to participate willingly. At first visit, all patients completed the Thai version ESS-CHAD and the questionnaires, including general demographic data (age, sex, height, weight, BMI), EDS symptoms, and sleep and other medical history. Children with risks of hypersomnia except OSA, for example, Sedative drugs, inadequate sleep, chronic disease e.g. heart disease, renal failure, liver failure, epilepsy, psychological problems, etc., were excluded from the study. Then, they were asked to do it again 1-2 hours later to check the test–retest reliability of the questionnaire. In order to check the discriminant validity of the ESS-CHAD, we included 26 patients with obstructive sleep apnea (OSA) confirmed by polysomnography. Participants underwent a standard PSG recording electroencephalogram, electro-oculogram, electromyogram, electrocardiogram, nasal pressure transducer, and thermistor for airflow measurement, thoracic and abdominal movement measurements, oxygen saturation monitoring, and a microphone for recording snoring sound. All polysomnographic data in our study were scored manually by experienced sleep technologists and reviewed by a board-certified specialist in sleep medicine. The

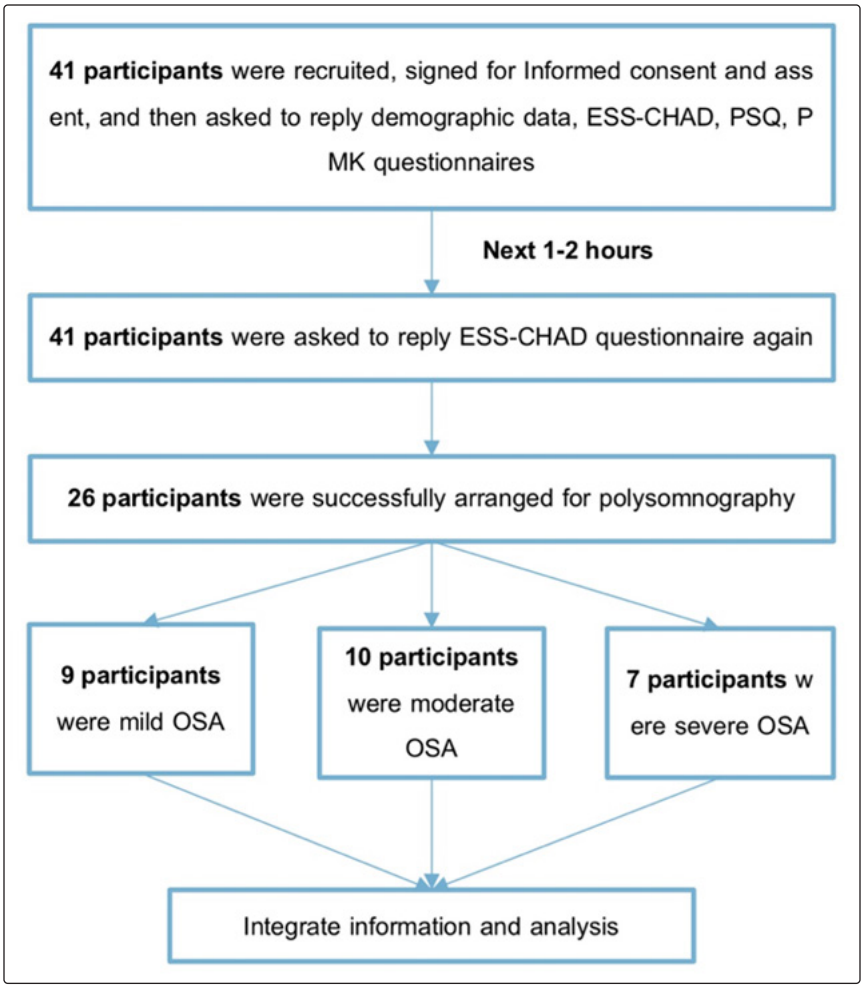

Figure 2 Flow diagram of methodologies for information gathering

AHI was calculated and used for diagnosis and classification of the disease's severity into three groups, namely, AHI of 1.5 to < 5 (mild OSA), AHI of 5 to 10 (moderate OSA), and AHI of > 10 (severe OSA). The results of PSG revealed 9 patients with mild OSA, 10 patients with moderate OSA, and 7 patients with severe OSA.

Statistical methods

To calculate the sample sizes for discriminant validity in this study, we used an intra-class correlation coefficient (ICC) of 0.79, 95% confidence interval of 1.96, rater of 2 and width of confidence of 0.17. Therefore, the initial estimated number of participants was 77. But due to the circumstance of pandemic COVID-19 infection, the number of patients in the hospital was reduced. So the estimated number of participants was re-calculated using an increased width of confidence.

This research was conducted using cross-sectional quantitative study design. For descriptive statistics, the continuous data will be presented in mean, standard deviation (SD), median, and range, and the categorical data will be presented in numbers and percentage. The statistics for reliability of ESS-CHAD was Cronbach's alpha coefficient, and the statistics for test-retest reliability of ESS-CHAD was Intra-class correlation coefficient. For the statistics for validity of ESS-CHAD, one-way ANOVA was used

to compare between the control group and the OSA group (mild, moderate, severe OSA), and Multiple comparison test was used to compare in details between the control group and the subgroup OSA. Pearson's correlation coefficients were used to determine the correlation between ESS-CHAD and PSG parameter. Statistical analysis was performed by using the STATA. Statistical significance was accepted at $p < 0.05$.

Results

Factors considered include gender, age group at the beginning of the study, body weight, height, and body mass index (BMI). The result showed that there were 41 subjects in total in this study, 31 males and 10 females, mean age of 10 years, mean body weight of 53.67 kg, mean height of 144.36 cm, and mean BMI of 24.62.

In the review of participants' comorbidities, the study showed that the most common comorbidity was overweight/obesity, followed by allergic rhinitis, and attention deficit hyperactivity disorder, respectively.

Table 1 Demographic data

Demographic data	n = 41
Sex, male (n, %)	31 (75.61%)
Age, mean±SD (years)	10.54±2.76
Body weight, mean±SD (kg)	53.67±22.55
Height, mean±SD (cm)	144.36±16.42
BMI, mean±SD (kg/m2)	24.62±6.57

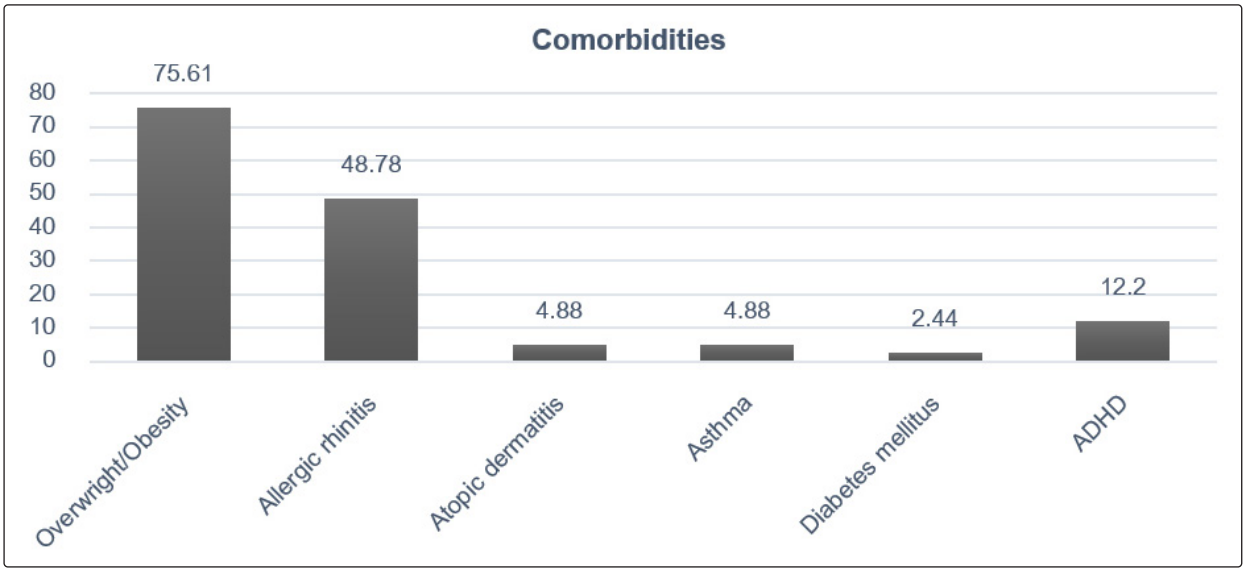

Figure 3 Percentage of comorbidities in all participants

Reliability

Cronbach's alpha coefficient for the ESS-CHAD Thai version in this study was 0.836 which indicated an excellent internal consistency. The test–retest reliability was done in 41 subjects. The ICC was 0.982 (95%CI: 0.967-0.991) with statistical significance (*p*-value < 0.05).

The ESS-CHAD scores between groups were evaluated. The results showed that the mean of ESS-CHAD scores in mild OSA was 3.33 with a standard deviation of 1.87, that in moderate OSA was 7.2 with a standard deviation of 5.65, and that in severe OSA was 9.86 with a standard deviation of 5.43.

There was a statistically significant between ESS-CHAD score and OSA (p-value <0.05). Also, there was a statistically significant difference between the mean of the ESS-CHAD scores of the mild OSA (3.33± 1.87) and the severe OSA patients (9.85± 5.43) (p < 0.05).

PSQ is a screening questionnaire for OSA, developed before ESS-CHAD. The cut-off score of ≥ 0.33 means highly possible OSA, and the score of < 0.33 means unlikely OSA. There was a statistically significant difference between the mean of the ESS-CHAD scores of the Pediatric Sleep Questionnairre (PSQ) negative patients (4.31±2.72) and the PSQ positive patients (7.36±5.74) (*p* < 0.05).

There was no statistically significant difference in the correlation between ESS-CHAD scores and polysomnogram parameters.

The ESS-CHAD score had the highest sensitivity of 70.59% and the highest specificity of 77.78% with a cut-off score ≥ 5.

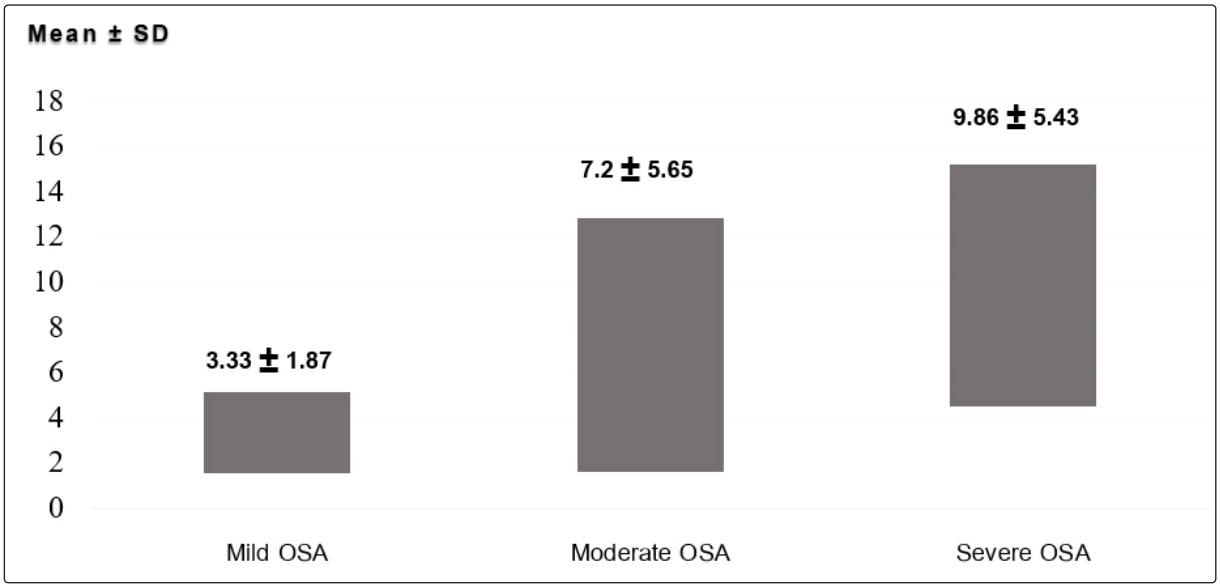

Figure 4 Comparisons of the ESS-CHAD scores between different groups

Table 2 Validity of ESS-CHAD compare to Polysomnogram (PSG) results

Validity-PSG	p-value*	*p*-value for multiple comparison test**		
		Mild vs Moderate	Mild vs Severe	Moderate vs Severe
ESS-CHAD score	0.031	0.246	0.031	0.768

Table 3 Validity of ESS-CHAD compare to Pediatric sleep questionnaire (PSQ)

Validity-PSQ	n	Mean	SD	*p*-value
ESS-CHAD score				0.026
PSQ-negative (score < 0.33)	13	4.31	2.72	
PSQ-positive (score ≥ 0.33)	28	7.36	5.74	

Table 4 The correlation between ESS-CHAD scores and polysomnographic parameters

	ESS-CHAD score		
	n	r	*p*-value
Arousal index (events/hr)	26	0.150	0.464
Stage NREM1 (%)	26	0.333	0.096
Stage NREM2 (%)	26	0.093	0.651
Stage NREM3 (%)	26	-0.257	0.206
Stage REM (%)	26	-0.005	0.981
Obstructive AHI (events/hr)	26	-0.063	0.759
Total AHI (events/hr)	26	0.212	0.299
SpO2 minimum (%)	26	-0.006	0.976
SpO2 average (%)	26	-0.274	0.175

**REM, rapid eye movement; NREM, non-REM; AHI, apnea-hypopnea index

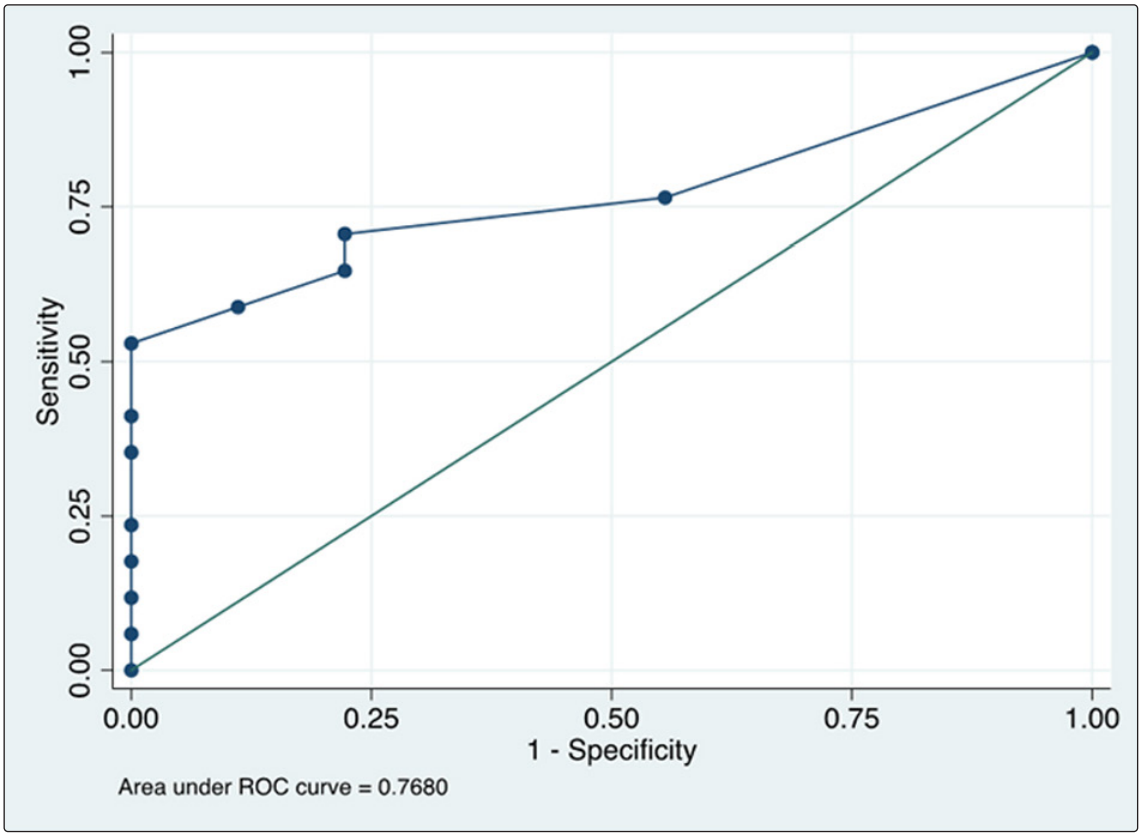

Figure 5 Sensitivity, Specificity, and ROC curve of the ESS-CHAD scores

Discussion

From the analysis in this study, the baseline characteristics of the study population show male predominantly participants with the mean age of 10 years and overweight/obesity as the most common comorbidity.

The reliability of the Thai version ESS-CHAD is analyzed and it shows excellent internal consistency (Cronbach's alpha coefficient = 0.836) and excellent reproducibility for the test–retest reliability (Intra-class correlation = 0.982, 95% CI ranging 0.967– 0.991) with statistical significance (p-value < 0.05). Compare to the study of the Thai version ESS in adults by Wish Banhiran[9], which had Cronbach's alpha coefficient = 0.87 and Intra-class correlation = 0.79, there is no difference in the reliability between the Thai version of ESS-CHAD and ESS. Compare to the Australian study of ESS-CHAD in excessive daytime sleepiness patients by Kitty C. Janssen[12], which had Cronbach's alpha coefficient = 0.73 and Intra-class correlation = 0.89, and the Australian study of ESS-CHAD in pediatric patients with narcolepsy with cataplexy by Y Grace Wang[11], which had Cronbach's alpha coefficient = 0.75 and Intra-class correlation = 0.755, the reliability of the Thai version ESS-CHAD is as excellent as the original language version.

The mean score of the ESS-CHAD obtained in the study population shows mild OSA (3.33± 1.87), moderate OSA (7.2±5.65), and severe OSA (9.85± 5.43). The association between ESS-CHAD scores and OSA patients is analysed and it shows that there is statistical significant association (p-value <0.05). Also, there is a statistically significant difference between the mean of the ESS-CHAD scores of the mild OSA and the severe OSA patients (p < 0.05). Subsequently, the ROC curve has been analyzed and it shows the highest sensitivity of 70.59% and the highest specificity of 77.78% with a cut-off score ≥ 5. Therefore, the Thai version ESS-CHAD is able to screen OSA patients with a cut-off value ≥ 5. In the study of ESS in excessive daytime sleepiness patients by Murray W. Johns in 1991[3],[4], the sensitivity was 93.5% and the specificity was 100% with a cut-off score > 10. When compared to our study, the sensitivity and specificity of ESS in adults are higher, which may be due to more suitable questions and more understandable individuals. According to the Australian study of the validation of ESS-CHAD in pediatric patients with narcolepsy with cataplexy by Y Grace Wang[11], the cut-off score > 10 suggests excessive daytime sleepiness, and the cut-off score ≥ 16 suggests a high level of excessive daytime sleepiness. Comparing with our study, the cut-off value for OSA is lower than the original language version, which may be due to lacking control group of primary snoring to compare with, and because the targeted study population between the three studies are different.

The association between ESS-CHAD scores and pediatric Sleep Questionnaire (PSQ), which is one of the screening questionnaires for OSA patients, is analysed and it shows that there is statisticalal significant association (p-value <0.05). From the study of Thai Version (ESS, SA-SDQ, and PSQI): Linguistic Validation, Reliability Analysis and Cut-Off Level to Determine Sleep Related Problems in Thai Population by Thanitpong Methipisit[10], the ESS was superior in predicting a good sleeper than SASDQ and PSQI in adults. In our study, the Thai version ESS-CHAD is able to screen OSA patients as well as the Thai version PSQ, and they correlate in the same direction. However, the analysis of the association between ESS-CHAD score and polysomnographic parameters shows no statistically significant difference, which may be due to small population.

The limitation of the study includes small sample size. Due to pandemic COVID-19 infection, there were several countermeasures launched during the year of 2020-2022, such as home isolation, social distancing, drug delivery to the patient's house, telemedicine, etc., which cause the reduction in the number of patients coming to Phramongkutklao hospital. So, the association requires further study with an increasing number of populations. Another limitation is age range. On account of the previous research on the original ESS-CHAD, it was conducted and used in population between age 7-17 years old and there was no other research that went below age 7. Also, the children's abilities in fully clarifying their own symptoms and understanding words and sentences are limited in young children, therefore, we only conducted this research in population between age 7-17 years old. So, we do not recommend using the questionnaire below age 7, since there have not enough solid evidences.

Conclusions

Our first Thai version of the ESS-CHAD showed an excellent internal consistency and test–retest reliability. It is able to discriminate between mild OSA patients and severe OSA patients with a cut-off value ≥ 5; however, it does not have a correlation with the polysomnogram parameters. Although, it is very useful for screening, it should not be used as a single tool to predict the OSA severity. We recommend the use of it in combination with a more comprehensive clinical evaluation.

Acknowledgement

This study was supported by Research Management Fund and Sleep disorder center, Department of Pediatrics, Phramongkutklao hospital, Thailand

Conflict of Interest

The authors declare that they have no conflict of interest.

Appendix

แบบประเมินอาการง่วงนอนของเด็กและวัยรุ่น ESS (CHAD)

ในช่วงสองถึงสามสัปดาห์ที่ผ่านมา คุณมีแนวโน้มที่จะง่วงสัปหงก หรือหลับขณะทำกิจกรรมต่างๆ ตามรายการดังต่อไปนี้มากน้อยแค่ไหน

แม้ว่าคุณจะไม่ได้ทำกิจกรรมดังกล่าวในช่วงนี้ แต่ลองคิดดูว่าแต่ละกิจกรรมจะส่งผลกับคุณอย่างไรบ้าง

ระบุตัวเลขที่ตรงกับคุณมากที่สุดระหว่างการทำกิจกรรมแต่ละอย่าง ตามระดับดังต่อไปนี้

0 = ไม่เคยรู้สึกง่วงเลย, 1 = มีโอกาสเล็กน้อยที่จะรู้สึกง่วง, 2 = มีโอกาสปานกลางที่จะรู้สึกง่วง, 3 = มีโอกาสสูงที่จะรู้สึกง่วง

กรุณาระบุคำตอบทุกข้อ

กิจกรรม	โอกาสที่จะรู้สึกง่วง			
	0	1	2	3
นั่งอ่านหนังสือ				
นั่งดูโทรทัศน์				
นั่งเรียนในห้องเรียนช่วงเช้า				
นั่งโดยสารในรถยนต์ต่อเนื่องเป็นเวลา 30 นาที				
เอนกายลงเพื่อพักผ่อนในช่วงบ่าย				
นั่งพูดคุยกับผู้อื่น				
นั่งคนเดียวหลังอาหารกลางวัน				
นั่งโดยสารในรถยนต์ขณะหยุดรอสัญญาณจราจรเป็นเวลา 2-3 นาที				

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