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The diagnostic accuracy of the Arabic version of the Pediatric Sleep Questionnaire for screening for pediatric sleep-related breathing disorders in Saudi children

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Abstract:

BACKGROUND: Pediatric Sleep Questionnaire (PSQ) is a valid, reliable tool for screening for sleep-related breathing disorders (SRBDs) translated into several languages since 2000. The diagnostic accuracy of an Arabic version of the PSQ has never been tested. Our aim was to translate the original version of PSQ into Arabic (Arabic-PSQ), validate it as a reliable screening tool, and compare it to the gold standard diagnostic method for SRBDs.

MATERIALS AND METHODS: This was a prospective longitudinal study of 54 children (2–14 years) who were to undergo polysomnography (PSG). SRBD was assessed by administering the Arabic version of PSQ to the parents of these children. The validity and reliability of the Arabic-PSQ were assessed. Data were analyzed using Stata 16. Correlation between with polysomnographic indices and PSQ scores, as well as measurement of the diagnostic accuracy were determined. Receiver operating characteristic analysis between the mean PSQ scores and binary PSG results was done and the area under curve (AUC) value was calculated.

RESULTS: Thirty-four (63%) children were diagnosed with obstructive sleep apnea by PSG (Apnea–Hypopnea Index [AHI] ≥1), 26 of whom were accurately identified with the Arabic-PSQ (76.5%). Arabic-PSQ showed comparable validity and reliability. Using a cutoff of 0.33, the score showed a significant correlation with AHI: *Rs:* 0.30 (*P* = 0.029). The sensitivity was 76.5%, the specificity was 50%, the positive predictive was 72.2%, the negative predictive value was 55.6%, the positive likelihood ratio was 1.63, and the negative likelihood ratio was 0.37.

CONCLUSIONS: The Arabic-PSQ is a valid tool for the screening of Arabic-speaking populations for SRBD. It is valuable for directing the diagnostic approach in a timely and cost-effective manner.

Keywords:

Obstructive sleep apnea, Pediatric Sleep Questionnaire, polysomnography, sleep disorders, validation

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Introduction

The burden of sleep-related breathing disorders (SRBDs) is recognized worldwide, with a prevalence of 2%–11% in the pediatric population,[1] SRBD involves a wide spectrum of clinical presentations based on the severity of airway restriction, ranging from primary snoring, upper airway resistance syndrome, obstructive hypoventilation, and obstructive sleep apnea (OSA) syndrome.[2] Both nighttime and daytime symptoms such as snoring, restless sleep, nocturnal enuresis, excessive sleepiness, hyperactivity, and school

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underperformance could be encountered.[3] OSA syndrome, considered the most severe form of SRBD in children, has a global prevalence of 0.7%–2%, and adenotonsillar hypertrophy is the most addressed comorbidity.[4] A linear relationship between the adenotonsillar size and Apnea–Hypopnea Index (AHI) has been reported.[5]

Although epidemiological studies regarding the prevalence of pediatric OSA in Saudi Arabia are lacking, it was estimated to be around 2.8%.[6] A cross-sectional study conducted on Saudi school children suggested that 21% are at high risk of developing SRBD.[7] This indicates that OSA, which might be underdiagnosed in children, results in growth failure, behavioral disorders like attention deficit-hyperactivity disorder and neurocognitive deficits with delays in learning.[4,8]

A detailed history and a comprehensive physical examination should be followed by polysomnography (PSG), the gold standard test to diagnose OSA, in selected patients. However, PSG is relatively time-consuming, expensive, and unavailable in resource-limited regions. This means there is the need for a simple, clinic-based, screening tool. In 2000, Chervin *et al*., developed the original version of Pediatric Sleep Questionnaire (PSQ) as a validated tool for the assessment of SRBD in children between 2 and 18 years of age, with a subscale argeting the diagnosis of OSA. It is a 22-item questionnaire with a sensitivity and specificity of 85% and 87%, respectively.[9] Given its advantages and utility, the PSQ-SRBD has been translated into several languages and validated. None of the studies done in Saudi Arabia performed an adequate diagnostic accuracy assessment, due perhaps to inadequate resources, difficult access to PSG and the conduct of the test on a large sample of patients being impractical.[10,11] Accordingly, in the present study, our aim was to translate the original version of into Arabic language (Arabic-PSQ), validate it as a reliable tool for screening children with SRDB, and assess its diagnostic accuracy by correlating its score to the PSG findings.

Materials and Methods

The study which was conducted from April 2021 to March 2022, employed a consecutive sampling to recruit all children aged 2–14 years referred to the pediatric sleep medicine clinic in a tertiary center in Riyadh, Saudi Arabia and were to undergo PSG to rule out SRDB. PSQ, a self-administered questionnaire for screening for obstructive SRBD, is composed of 22 closed-ended questions in four groups based on such prominent symptoms as snoring, daytime sleepiness and inattentive or hyperactive behavior, and others. The response options "yes," "no," or "don't know" were assigned numerical values of 1, 0, or missing, respectively. The proportion of questions answered affirmatively is calculated. Unanswered questions or items responded to as "don't know" were subtracted from the denominator when calculating the proportion. A cutoff point of 0.33 was established.[9] Consequently, the values exceeding 0.33 were deemed positive, potentially indicating the presence of pediatric obstructive sleep-disordered breathing. Approval of translating SRBD scale into Arabic was obtained from the original author of the questionnaire "Chervin RD" and University of Michigan, USA. Ethical approval was obtained from the Institutional Review Board (IRB) vide Letter No. IRB-2021-01-098 dated 10/03/2021, and written informed consent was taken from all participants in the study.

The original version of the questionnaire underwent translation and cross-cultural adaptation in accordance with the recommended procedure outlined by Beaton *et al*.[12] The initial step involved the independent forward translation of the questionnaire from English to Arabic by two separate translators. The two versions were compared for any discrepancies or ambiguities and a common Arabic version generated. Back-translation was done independently by two translators who were blind to the original questionnaire, then an expert committee compared the two versions to the original questionnaire. A subgroup of thirty parents filled the Arabic-PSQ twice, in an interval of 4 weeks, to calculate the test-retest reliability and confirm the long-term stability of responses before the final translated version was made. The investigators collected information on the clarity of the questionnaire. For cultural relevance, question C14 was adapted to Arabic context and the phrase changed from "Is 'on the go' or often acts as if 'driven by a motor'" to "suffers from hyperactivity." The final Arabic-PSQ was then reviewed and generated.

Paper-based questionnaires were distributed to parents on the same visit of PSG study. Informed consent was attached to be signed by the parent, after an explanation and the purpose and nature of the study were given. Completed questionnaires were sent directly and anonymously to the primary author for analysis. The data were copied to a password-protected Excel sheet accessible only to the primary investigator, and descriptive statistics were calculated.

Overnight PSG was performed on the entire study sample in a sleep laboratory using a computerized system. The PSG involved recording sleep stages through electroencephalographic, electromyographic, and electro-oculographic leads. The criteria outlined in the American Sleep Disorders Association Task Force report were used to analyze the respiratory events, sleep

architecture, and identification of arousals.[13] Central, obstructive, and mixed apneas and hypopneas were defined as per the current recommendations.[14] OSA was defined as an AHI ≥1. PSG analysis was conducted with no knowledge of the results from the PSQ, and a statistical comparison was made between the total PSQ score of each patient and their PSG indices.

Analysis was done using the StataCorp. 2019. Stata Statistical Software: Release 16. College Station, TX, USA: StataCorp LLC. The psychometric properties were assessed, including the calculation of internal consistency for each scale using Cronbach's alpha. Adequate reliability was indicated by a value >0.7, moderate reliability fell within the range of 0.5–0.7, fair reliability was within 0.2–0.4, and low reliability was suggested by a value <0.2, aligning with the criteria set by Nunnally and Bernstein for group comparisons.[15] Long-term stability of item responses was evaluated by test–retest reliability using Spearman's correlation coefficient. $P < 0.05$ indicated statistical significance. To assess the diagnostic accuracy, sensitivity, specificity, and predictive values were determined. Receiver operating characteristic analysis between the mean PSQ scores and binary PSG results was done and the area under curve (AUC) value was calculated.

Results

This study of a total of 54 participants who completed both the PSQ and PSG have their characteristics listed in Table 1. The majority of cases were male (40; 74%). The mean age was 7.1 ± 3.8 years. About 24% had body mass index (BMI) above the 90th percentile and 66% had comorbidities. Thirty-four (63%) children were diagnosed with OSA by PSG (AHI ≥1). The mean Arabic-PSQ score was 0.41.

The PSQ cutoff value of 0.33 was applied to the entire sample. Table 2 shows the correlations between PSG indices and PSQ score. The PSQ score statistically analyzed to the AHI revealed a significant correlation: *Rs*: 0.30 ($P = 0.029$). The Spearman's correlation between a set of PSG indices and PSQ score was established. The correlations between the PSQ score with the arousal index, baseline SpO_2, and baseline CO_2 were 0.09 ($P = 0.497$), −0.18 ($P = 0.194$), and 0.29 ($P = 0.032$), respectively.

Spearman's correlation coefficients for the test–retest analysis showed excellent reliability, with values of 0.982 for the total SRBD scale ($P < 0.05$), 0.995 for the snoring scale ($P < 0.05$), 0.942 for the daytime sleepiness domain ($P < 0.05$), and 0.960 for the hyperactivity domain ($P < 0.05$). As detailed in Table 3, most of the Arabic-PSQ items were significantly correlated with the PSQ global score. Items that did not show statistical significance are "unrefreshed in the morning" ($P = 0.133$), "delayed growth" ($P = 0.154$) and "obesity" ($P = 0.386$).

The Arabic-PSQ scale showed an internal consistency, with a Cronbach's alpha coefficient of 0.83 for all items, 0.86 for the snoring domain, 0.81 for the daytime sleepiness domain, 0.77 for the hyperactivity domain, and 0.34 for other symptoms [Table 4].

Table 1: Characteristics of Saudi children aged 2-14 years undergoing polysomnography (*n*=54)

Characteristics	*N* (%)
Sex	
Male	40 (74.0)
Female	14 (26.0)
Age	
Mean±SD	7.1±3.8
Median	7.5
Range	12 (2–14)
BMI percentile	
≤25th	11 (20.4)
25–50th	12 (22.2)
50–90th	18 (33.3)
>90th	13 (24.1)
Comorbidities	
Hyperactive airway disease	13 (24.1)
Medical other than respiratory diseases	17 (31.5)
Neuromuscular disease	5 (9.3)
Intellectual disability	5 (9.3)
No comorbidity	18 (33.3)
PSG results (AHI)	
Negative (<1)	20 (37.0)
Mild (1–5)	23 (42.6)
Moderate (>5–10)	8 (14.8)
Severe (>10)	3 (5.6)
Arabic-PSQ score	
Identified cases	26 (76.5*)
Mean±SD	0.41±0.23
Minimum	0.0
Maximum	0.91

*The number of identified cases by the questionnaire out of all true positives whom were detected by PSG. PSG=Polysomnography, BMI=Body mass index, AHI=Apnea–Hypopnea Index, PSQ=Pediatric Sleep Questionnaire, SD=Standard deviation, Arabic-PSQ=PSQ into Arabic

Table 2: Correlations between polysomnography indices and Pediatric Sleep Questionnaire score

	r	*P*-value
Total sleep time	−0.00	0.968
Sleep latency	−0.20	0.15
Sleep efficiency	0.05	0.693
AHI	0.30	0.029
Obstructive/mixed events	0.32	0.021
Arousal index	0.09	0.497
Baseline SpO_2	−0.18	0.194
Minimum SpO_2	−0.16	0.25
Baseline CO_2	0.29	0.032
Maximum CO_2	0.30	0.028

AHI=Apnea–Hypopnea Index

With a cutoff value of 0.33, the Arabic-PSQ score showed a significant correlation with the AHI, which was set at a cutoff point of AHI > 1 indicating OSA-positive case. Of high-risk participants (AHI > 1), 26/34 were accurately identified by PSQ (76.5%). The results showed that the sensitivity of the Arabic-PSQ to OSA was 76.5% (95% confidence interval [CI] = 65.2%–87.8%), the specificity was 50% (95% CI = 36.66%–63.34%), the positive predictive was 72.2% (95% CI = 60.3%–84.2%), and the negative predictive value was 55.6% (95% CI = 42.3%–68.8%) [Table 5]. The AUC was estimated at 0.64 [Figure 1].

Table 3: Distribution of Pediatric Sleep Questionnaire items and their correlation with Pediatric Sleep Questionnaire global score

Items	"Yes" response *N* (%)	Spearman's correlation	
		r	*P*-value
Snoring domain			
Usually snores	26 (48.2)	0.6	<0.001
Always snores	22 (40.7)	0.5	<0.001
Snores loudly	22 (41.5)	0.7	<0.001
Heavy breathing	27 (50.9)	0.6	<0.001
Trouble breathing	28 (51.9)	0.5	<0.001
Observed apneas	20 (38.5)	0.3	0.015
Mouth open during day	31 (57.4)	0.6	<0.001
Dry mouth on awakening	38 (70.4)	0.5	<0.001
Daytime sleepiness domain			
Unrefreshed in morning	26 (48.2)	0.2	0.133
Problem with sleepiness	16 (29.6)	0.5	<0.001
Sleepy per teacher	14 (34.2)	0.5	<0.001
Hard to wake-up	24 (45.3)	0.4	0.003
Inattention/hyperactivity domain			
Does not listen	21 (41.2)	0.6	<0.001
Difficulty organizing	24 (47.1)	0.7	<0.001
Easily distracted	20 (40.0)	0.5	<0.001
Fidgets	22 (42.3)	0.6	<0.001
Suffers from hyperactivity*	16 (30.2)	0.4	0.004
Interrupts	15 (29.4)	0.3	0.017
Other symptoms			
Nocturnal enuresis	16 (31.4)	0.4	0.009
Morning headache	11 (23.9)	0.5	0.002
Delayed growth	9 (17.7)	0.2	0.154
Obesity	11 (20.8)	0.1	0.386

*This item was originally stated as "on the go/driven by motor" but changed after cultural adaptation of the questionnaire

Table 4: Cronbach's alpha of the global score and the subdomains of Arabic version of PSQ

Arabic-PSQ scale	Cronbach's alpha
All items	0.83
Snoring	0.86
Daytime sleepiness	0.81
Inattention/hyperactivity	0.77
Other symptoms	0.34

Discussion

This study is a modest contribution to the ongoing efforts related to the translation and cross-cultural adaptation of PSQ, the aim of which is to enhance its efficacy as a screening instrument for Arabic-speaking population. The data were prospectively collected from children referred for PSG assessment, and a diagnosis of OSA was confirmed in about 63%. The Arabic-PSQ demonstrated a significant correlation with PSG results when AHI ≥1 was used as a cutoff with good internal consistency, excellent reliability, and long-term stability of questionnaire. OSA was confirmed by PSG in about 63%. Of the patients who were diagnosed with OSA, about 72% were accurately identified as high risk by the PSQ.

Eight patients diagnosed by PSG but were not detected by PSQ; seven of these patients had mild OSA with obstructive indices <1 with no gas exchange abnormality. The remaining patient was a known case of down syndrome and had moderate OSA (AHI = 7.3). This might suggest that the sensitivity of PSQ increases with the severity of the disease. Our findings encourage the application of the Arabic-PSQ in the clinical setting to screen for SRDB, showcasing a sensitivity and specificity of 76.5% and 50%, respectively.

SRBD can stem from either a combination of such physiological factors as BMI, or anatomical factors of enlarged adenoids, narrow arches, and a high soft palate. A study indicated that the likelihood of SRBD increases six-fold in children experiencing noncontinuous sleep, five-fold in those exhibiting mouth breathing, and over three-fold in those who snore.[16] PSG serves as the gold standard diagnostic test for SRBD. Nevertheless, owing to concerns of high cost, the shortage of pediatric sleep centers and the burden of disease, universal efforts have been made to assess screening methods and reserve PSG to high-risk patients.[1]

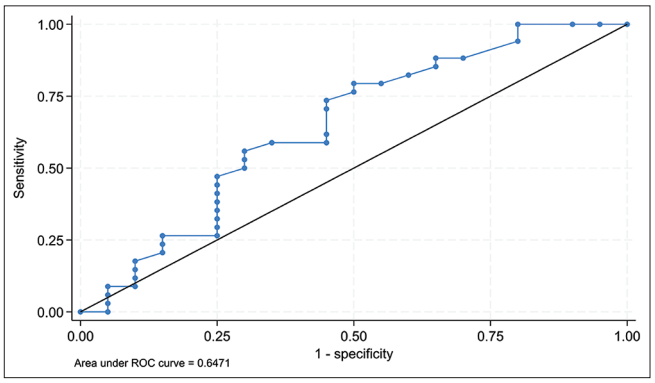

Figure 1: Receiver operating characteristic analysis between mean Pediatric Sleep Questionnaire scores and polysomnography results. ROC = Receiver operating characteristic

Table 5: Diagnostic value for the Pediatric Sleep Questionnaire when using 0.33 as a cutoff point

PSQ	PSG (AHI ≥1)		Total
	No	Yes	
No	10	8	18
Yes	10	26	36
Total	20	34	54
Sensitivity, Precent (95% CI for %)	76.5% (65.2%–87.8%)		
Specificity, Precent (95% CI for %)	50.0% (36.7%–63.3%)		
Positive predictive value, Precent (95% CI for %)	72.2% (60.28%–84.2%)		
Negative predictive value, Precent (95% CI for %)	55.6% (42.3%–68.8%)		

PSQ=Pediatric Sleep Questionnaire, PSG=Polysomnography, AHI=Apnea–Hypopnea Index, CI=Confidence interval

The role of PSQ extends from being a preliminary tool for diagnosis to addressing a constellation of symptoms that can be improved by treating breathing disorders. It can also encourage effective multidisciplinary collaboration between otolaryngologists, pediatric physicians, dentists, and psychologists. In addition, parental introspection can help in understanding the disease, thereby promoting their active involvement in the treatment process.

The original version of PSQ has a sensitivity of 81% and a specificity of 87%.[9] Numerous studies have translated the questionnaire into different languages, including French, Spanish, Chinese, Thai, Malay, Indian, Portuguese, Danish, and Turkish.[2,4,8,17-22] The internal consistency and reliability of the translated versions were found to be comparable to our results. Utilizing a cutoff of 0.33, the sensitivity of PSQ in the literature ranged between 62% and 88.2%.[4,9,18,23-24] The specificity of the PSQ varied widely among studies, between 52% and 87%. In our study, the AUC was 0.64, which is consistent with the values reported in the literature (0.57–0.69).[23,25] The diagnostic accuracy of our translated version appears to be comparable to the original and other validated versions of the questionnaire.

Some studies focused on special groups of patients and merit comment. Ehsan *et al*., found PSQ to be a reliable screening tool for OSA in asthmatic patients, with acceptable sensitivity.[23] Another data on primary snorers showed that PSQ helped in identifying 89% of children with OSA with excellent negative likelihood ratio (0.261).[4] It is worth mentioning, however, that the specificity was found to be as low as 36.9% in asthmatic patients and 17.1% in snorers.[23,24] The specificity did not increase even after integrating the score with the clinical findings. This inconsistency may be attributed to sampling bias. Yet, the concern arises from the fact that snorers and patients with comorbidities constitute the population that will benefit the most from this screening tool; therefore, a very low specificity raises the potential for misguided interpretation of the PSQ. Our data encompassed all children presenting with clinical symptoms of sleep disordered breathing in which OSA was one of the top differential diagnoses and referred for PSG. The questionnaire showed a specificity of 50% and a good negative likelihood ratio (0.55), both of which are considered acceptable for a screening tool. Overall, a trade-off between sensitivity and specificity is expected, and the careful interpretation of the PSQ in the preliminary screening stage of OSA can serve as a helpful guide for the evaluation of OSA, especially for high-risk patients.

The burden of SRDB in Saudi Arabia cannot be overlooked. The estimated percentage of pediatric children at high risk of developing SRBD is 13%–21%, with a higher risk observed in children aged 6–9 years.[25,26] Clinical factors strongly associated with SRBD include habitual snoring, witnessed apnea, mouth breathing, bedwetting, and being overweight. To our knowledge, only two studies have aimed at providing a validated Arabic version of the PSQ and have shown excellent internal consistency.[10,11] Both studies were conducted on Saudi Arabian populations and were published recently (2023). The translated version by Almutairi *et al*., was tested and validated on 72 participants divided into two groups based on the probability of OSA.[11] Their version was found to be reliable, and Cronbach's α values were 0.805 for the total score, 0.799 for the snoring domain, 0.690 for the sleepiness domain, and 0.711 for the behavioral domain. The sensitivity and specificity of PSQ were determined after PSG was performed on only 15 participants and were 79.6% and 43.5%, respectively, using a cutoff score of 0.33. In contrast to our study, however, the authors did not find a significant correlation between the AHI and the questionnaire total score, possibly because of their small sample. The second study was done by Mazi on patients with adenotonsillar hypertrophy and/or obesity.[10] It showed superior internal consistency, with a Cronbach's α value of 0.946 for the total score, 0.924 for the snoring domain, 0.762 for the sleepiness domain, and 0.820 for the behavior domain. There was a statistically significant correlation between items within each subscale, yet some items were found to cross-correlate with more than one subscale. Our study demonstrated comparable internal consistency to both reports.

This study has several limitations. First, it was a single-center study with a low number of participants. In this prospective study, we chose to perform PSG on all study participants and collect responses on the Arabic-PSQ at the same PSG visit, for a trade-off between the number of included participants and the precision of result interpretation. Second, we did not conduct a subanalysis of the results based on age, gender, and BMI. In the study by Chervin *et al*., age was not found to be

a significant factor, while female gender was associated with higher behavioral scores.[9] Ehsan *et al*., found a strong correlation between obstructive index and PSQ in obese children and those under 13 years of age.[23] Third, we did not evaluate the validity of the questionnaire in assessing clinical improvement following surgery, which can determine whether the scale can realistically reflect the clinical change and be used to evaluate the success of treatment. Almutairi *et al*.,[11] found a significant difference between the mean score before (0.464) and after (0.185) surgery, with greater marked improvement in the breathing domain. This is probably because behavioral problems do not improve directly after surgery when the obstruction is resolved. Fourth, this study had no control group. The diagnostic accuracy of the questionnaire was not tested on children who had no indication of undergoing PSG test to correlate with clinical judgment. This limitation was also observed in the original validation study.[9] We strongly recommend testing the Arabic-PSQ on a larger group of Saudi and non-Saudi Arabian populations, using subscale and subgroup analyses, and conducting follow-up responses to correlate preoperative and postoperative findings.

Conclusion

The Arabic-PSQ is a valid tool that can be used on the Arabic-speaking population to screen for SRBD. It showed a significant correlation with PSG findings when AHI > 1 was used as a cutoff, with a sensitivity of 76.5% and a specificity of 50%. By integrating this screening tool into clinical practice, the decision to undergo PSG would be further justified after approaching the patient in a timely and cost-effective manner. Further studies to consolidate this culturally adapted version are highly encouraged.

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Conflicts of interest
There are no conflicts of interest.

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