



# Is the Pediatric Sleep Questionnaire sensitive for sleep-disordered breathing in children with complex chronic disease?

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## Abstract

**Purpose** Sleep-disordered breathing (SDB) is a disease defined by breathing or breathing irregularities while asleep. The current study examines the association between results of polysomnography (PSG) and the Pediatric Sleep Questionnaire (PSQ), and the specificity and sensitivity of the PSQ for obstructive sleep apnea (OSA) in patients with chronic illnesses.

**Methods** Demographic and clinical attributes, in addition to PSQ and PSG outcomes were examined retrospectively among patients who underwent polysomnography (PSG) at our facility between 2012 and 2021.

**Results** Of 745 patients included in the study, 462 (62%) were male. The median age was 81 months (34–151 months). 117 of the patients (15/8%) had chronic lung disease, and 80 (10.7%) had cerebral palsy. The most common indications for PSG were symptoms of OSA ( $n=426$ ; 57.1%). According to obstructive apnea-hypopnea index (AHI), 361 patients (48.5%) had normal PSG. The median PSQ score was 0.40 (0.22–0.57). The sensitivity and specificity of the PSQ were 71.8% and 40.4%, respectively, for individuals aged 2 to 18 years. Among the disease subgroups, the cerebral palsy group had the highest sensitivity of PSQ (88.8%) for diagnosis of OSA.

**Conclusion** Questionnaires for evaluating SDB are not sensitive or specific for identification of OSA in children with chronic conditions, and PSG remains the best method.

**Keywords** Children · Chronic disease · Pediatric Sleep Questionnaire · Polysomnography

## Abbreviations

SDB	Sleep-disordered breathing	BiPAP	Bilevel positive airway pressure
OSAS	Obstructive sleep apnea syndrome	BMI	Body mass index
CSA	Central sleep apnea	PWS	Prader-Willi syndrome
ICSD-3	International Classification of Sleep Disorders – Third Edition	REM	Rapid eye movement
PSG	Polysomnography	WASO	Waking up after sleep onset
PSQ	Pediatric Sleep Questionnaire	SL	Sleep latency
AASM	American Academy of Sleep Medicine	SE	Sleep efficiency
TST	Total sleep time	NMD	Neuromuscular disease
AI	Apnea index	PPV	Positive predictive value
HI	Hypopnea index	NPV	Negative predictive value
AHI	Apnea-hypopnea index	CI	Confidence interval
oAHI	Obstructive apnea–hypopnea index	ENT	Ear, nose, and throat
CAI	Central apnea index	NIV	Non-invasive ventilation
ODI	Oxygen desaturation index	ERS	European Respiratory Society
PAP	Positive airway pressure	DS	Down syndrome
CPAP	Continuous positive airway pressure	AT	Adenotonsillectomy

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## Introduction

Sleep-disordered breathing (SDB) is a disease defined by breathing or breathing irregularities while asleep. SDB includes sleep-related hypoventilation, hypoxemia disorders, central sleep apnea (CSA), and obstructive sleep apnea (OSA) [1]. Recurring bouts of partial or total obstruction of the respiratory tract impair ventilation and cause OSA. Diseases in the OSA spectrum comprise primary snoring, upper airway resistance syndrome, and OSA. The frequency of OSA among children is approximately 2–4% [2]. The third edition of the International Classification of Sleep Disorders (ICSD-3) defines CSA with a brief reduction in airflow or cessation of breathing due to a lack of or reduced respiratory effort [3]. The prevalence ranges between 4 and 6% in children with SDB. Children with underlying illnesses like anatomical brain and brainstem abnormalities, neurogenetic disorders, prematurity, gastroesophageal reflux disease, obesity, and hypothyroidism are more likely to develop CSA [4]. SDB, left untreated, can result in serious side effects such as pulmonary hypertension, cor pulmonale, and failure to thrive. In addition, SDB may cause learning and behavioral problems by affecting children's cognitive functions. It is essential to diagnose and treat SDB in a timely fashion due to its significant effects on health and the global healthcare burden [4].

Overnight polysomnography (PSG) has been accepted as the most precise diagnostic method in children with a high suspicion of SDB [2]. However, due to its high cost, lack of pediatric sleep laboratories, and lack of experienced healthcare personnel, several questionnaires have been developed as screening tools to evaluate patients at elevated risk of OSA. Screening questionnaires are straightforward and inexpensive tools, and it was recommended that they may be a tool for the diagnostic assessment of children with indicators or symptoms of OSA, especially in low-resource settings [5, 6]. The Pediatric Sleep Questionnaire (PSQ) is currently commonly used to detect moderate to serious OSA in healthy children aged 2 to 18 years [7]. However, the PSQ has also been used in various disease groups in recent years with varying success [6, 8].

The primary objective of this study was to assess the outcome of children with SDB symptoms who had been examined in our sleep laboratory and examine their demographic, clinical, and polysomnographic data. The secondary purposes were to assess the correlation between PSQ and PSG results and to evaluate the sensitivity and specificity of the PSQ in patients with underlying diseases.

## Materials and methods

### Participants and study design

The retrospective study was carried out at the Pediatric Pulmonology Division of Marmara University between January 2012 and January 2021. Patients performing sleep studies between the ages of 0 and 21 were included. Clinical and demographic details regarding the patients were taken from the medical records. The Marmara University ethics committee approved the research protocol (07.05.2021–09.2021.659).

Data collected included baseline demographics (age, gender, height, weight), underlying diseases, symptoms related to SDB such as snoring, mouth breathing, venous blood gas carbon dioxide level, indications for PSG, PSQ, PSG findings, and treatment modalities.

### Pediatric Sleep Questionnaire

The PSQ is used to assess breathing disorders associated with sleep-related symptoms. Twenty-two questions on this parent-reported survey ask about snoring, observed apneas, difficulty breathing while sleeping, and other characteristics of OSA [5]. “Yes” equals 1, “no” equals 0, and “don't know” is regarded as a missing response. The average response falls between 0 and 1. To calculate the score, the total number of “yes” answers is divided by the total amount of questions. For pediatric SDB, a threshold of 0.33 is used to determine elevated risk [5].

### Polysomnography

Patients underwent a standard in-laboratory, overnight video PSG based on recommendations from the American Academy of Sleep Medicine (AASM) [2]. Doctors assessed all PSGs after being scored by skilled pediatric sleep technicians. The total number of obstructive, central, and mixed apneas and hypopneas divided by the number of hours of uninterrupted sleep was called the apnea–hypopnea index (AHI). The total number of obstructive apneic and hypopneas events per hour of sleep was called obstructive AHI (oAHI). The severity of OSA was evaluated according to the oAHI of the patients [9]: mild if  $\text{oAHI} \leq 5$  events·h<sup>-1</sup> but  $> 1$  events·h<sup>-1</sup>, moderate if it is  $> 5$  events·h<sup>-1</sup> but  $\leq 10$  events·h<sup>-1</sup>, and severe if it was  $> 10$  events·h<sup>-1</sup> [9, 10]. Central apnea was diagnosed if CAI is  $> 5$  events·h<sup>-1</sup> [9, 10].

## Statistical analysis

The statistical analyses utilized the Statistical Package for the Social Sciences (version 16.0; SPSS Inc.; Chicago, IL, USA). For information that did not distribute normally, the Mann–Whitney *U* and Kruskal–Wallis tests were used to compare the study groups and descriptive statistical methods used median with interquartile range. To compare more than two groups, we used the Kruskal–Wallis test. The Mann–Whitney *U* test was done and compared with the Bonferroni correction in pairwise comparisons. Spearman’s rank correlation was used to assess bivariate correlations, and the results were displayed as correlation coefficients. A chi-square test was used to compare variations in proportions between two independent groups. A *p*-value of <0.05 was regarded as statistically significant.

## Results

Polysomnography was performed for 745 patients in the sleep laboratory within the study period. Table 1 shows information about the patients’ demographic features.

The most common indications for PSG were symptoms of OSAS (*n*: 426, 57.1%), including snoring (54.7%), mouth breathing (51.4%), and witnessed apneas (47.5%). Polysomnography was indicated for pCO<sub>2</sub> retention in venous blood gas in 181 (24.3%) patients and evaluation prior to growth hormone treatment in patients with Prader-Willi syndrome (PWS) in 43 (5.8%) patients. Thirty-seven (5%) patients had PSG for pre-decannulation evaluation, 29 (3.9%) patients had PSG in the assessment

of pulmonary hypertension, 23 (3.1%) patients were evaluated for desaturations, and 6 (0.8%) patients were studied for PAP start.

According to oAHI, 361 patients (48.5%) had normal PSG, 195 patients (26.2%) had mild, 68 patients (9.1%) had moderate, and 121 patients (16.2%) had severe OSA. Central apnea was diagnosed in 75 (10.1%) patients. Table 2 displays respiratory and sleep parameters. The median oAHI of patients with suspected SDB, according to PSQ, was 1.25 events-h- (0.1–7.3). When we contrasted the oAHI and PSQ scores, we observed a favorable correlation between the PSQ and oAHI values ( $r=0.103$ ,  $p<0.02$ ).

Table 3 compares the underlying illnesses with age, respiratory parameters, and PSQ. Age, AHI, CAI, oAHI, and PSQ differed significantly from disease subgroups ( $p<0.05$ ). Patients with NMD were considerably older than those with genetic-metabolic diseases ( $p<0.001$ ). However, their AHI values were substantially lower than those with genetic-metabolic disorders ( $p<0.001$ ). Compared to patients with chronic lung disease, patients with genetic-metabolic diseases had significantly higher oAHIs ( $p<0.01$ ). The CAI of patients with NMD was markedly higher than those with endocrine disorders ( $p<0.005$ ). The PSQ scores of patients with cardiological diseases were notably lower than those of obese patients ( $p<0.007$ ).

For every different disease group in children aged 2 to 18, the PSQ’s sensitivity, specificity, and other parameters were calculated (Table 4). The outcomes for the groups with upper airway issues, cardiovascular issues, and obesity were not in the table due to a lack of PSQ data in those groups.

**Table 1** Demographic data of the study group

Age (month), median (25–75p)	81 (34–151)
Male, <i>n</i> (%)	462 (62)
Body mass index (BMI), median (25–75p)	16.4 (13.8–20.7)
Blood venous gas PCO <sub>2</sub> (mmHg), median (25–75p)	45 (39–50)
PSQ, median (25–75p)	0.40 (0.22–0.57)
The underlying medical conditions of the patients	
*Chronic lung disease, <i>n</i> (%)	117 (15.8)
*Genetic-metabolic syndrome, <i>n</i> (%)	98 (13.2)
*Neuromuscular disease, <i>n</i> (%)	87 (11.7)
*Endocrine diseases, <i>n</i> (%)	82 (11)
*Cerebral palsy, <i>n</i> (%)	80 (10.7)
*Otherwise healthy, <i>n</i> (%)	79 (10.6)
*Upper airway problems, <i>n</i> (%)	63 (8.5)
*Cardiological diseases, <i>n</i> (%)	45 (6)
*Obesity, <i>n</i> (%)	30 (4)
*Others (chest deformities, scoliosis, diaphragmatic eventration, congenital central hypoventilation syndrome, spina bifida + hydrocephalus), <i>n</i> (%)	64 (8.5)

**Table 2** Respiratory and sleep parameters from PSG

AHI, median (25–75p)	2.3 (0.65–8.30)
CAI, median (25–75p)	0.5 (0–1.60)
OAHI, median (25–75p)	1 (0.05–5.11)
ODI, median (25–75p)	4.5 (1.40–13.65)
Minimum O <sub>2</sub> saturation, median (25–75p)	89 (83–93)
N1 ratio, median (25–75p)	4.55 (2.1–9.6)
N2 ratio, median (25–75p)	46.3 (31.97–57.50)
N3 ratio, median (25–75p)	33 (20.9–45.10)
REM ratio, median (25–75p)	5.75 (0–12.80)
TST, median (25–75p)	336.85 (169.87–410.85)
WASO, median (25–75p)	25 (10.5–50.5)
SL, median (25–75p)	17 (6.85–36)
SE, median (25–75p)	83 (70–91)
REM latency, median (25–75p)	86.5 (30.87–187.12)

AHI, apnea–hypopnea index; CAI, central apnea index; OAHI, obstructive apnea–hypopnea index; ODI, oxygen desaturation index; REM, rapid eye movement sleep; TST, total sleep time; WASO, waking up after sleep onset; SL, sleep latency; SE, sleep efficiency

Three hundred four (40.8%) of 745 patients were treated after PSG. Table 5 lists the available treatment implemented for them. Nineteen (6.2%) of the 37 tracheostomy patients were decannulated. Ear, nose, and throat (ENT) analysis was recommended for upper airway evaluation in all patients with oAHI > 1 events·h<sup>-1</sup>. Since two patients (0.6%) were morbidly obese, follow-up was planned with an ENT examination and weight loss recommendation.

## Discussion

This large, retrospective cohort study revealed that OSA was notably greater among children with accompanying conditions like genetic-metabolic, neuromuscular, and chronic lung diseases. In addition, the PSQ score was increased in all patient groups and the underlying subgroup of diseases except for the cardiological conditions.

The PSQ is among the best tools for predicting OSA since it is among the validated questionnaires for assessing SDB in healthy children in resource-limited settings [11]. It is an effective screening tool, mainly due to the difficulty of accessing sleep laboratories, and to help alleviate long waiting lists for PSG. In our research, for the patients 2–18 years of age, the sensitivity and specificity of PSQ were 71.8% and 40.4%, respectively. Among the disease subgroups, the group in which the PSQ had the highest sensitivity (88.8%) was the cerebral palsy group. However, the specificity was 30.7% in the same group of patients with CP who had experienced more significant sleep

**Table 3** Relationship between AHI-PSQ by disease subgroups

Median (25p–75p)	All disease (n: 745)	Chronic lung disease (n: 117)	Genetic-metabolic syndrome (n: 98)	Neuromuscular disease (NMD) (n: 87)	Endocrine diseases (n: 82)	Cerebral palsy (n: 80)	Healthy (n: 79)	Upper airway problems (n: 63)	Cardiological diseases (n: 45)	Obesity (n: 30)	Others (n: 64)	P*
Age (month)	78.50 (33–150)	97 (36–166)	58 (16.50–112.50)	114(53.25–165)	47 (17–130.50)	69 (35.50–131.50)	77.50 (39–148.25)	59 (33–133)	96 (50–136)	114 (76–166)	87 (42–172)	p<0.001
AHI	2.3 (0.67–8.32)	1.45 (0.20–3.10)	5.95 (2–15.27)	1.20 (0.40–3.40)	3.41 (1.17–9.72)	2.15 (0.60–8.67)	1.50 (0.40–6.40)	2.40 (0.80–12.30)	1.90 (0.55–5.15)	3 (0.15–8.57)	2.4 (0.54–13.87)	p<0.001
OAHI	1 (0.07–5.14)	0.35 (0–1.97)	3.95 (0.85–12.90)	0.60 (0–2.40)	1.17 (0.27–6)	1.35 (0.12–4.60)	0.60 (0–2.60)	1.50 (0.10–8.90)	1.10 (0.30–3.95)	1.6 (0–7)	1.15 (0–11.72)	p<0.001
CAI	0.5 (0–1.61)	0.5 (0–1.4)	0.62 (0–2)	0.2 (0–0.9)	1.27 (0.2–3.2)	0.50 (0–1.45)	0.4 (0–1.80)	0.60 (0–2)	0.35 (0–1.70)	0.8 (0–2.12)	0.47 (0–1.30)	p<0.005
PSQ	0.40 (0.22–0.57)	0.37 (0.21–0.60)	0.44 (0.22–0.64)	0.33 (0.16–0.47)	0.33 (0.22–0.56)	0.47 (0.29–0.60)	0.38 (0.25–0.60)	0.45 (0.26–0.59)	0.31(0.18–0.47)	0.50 (0.42–0.68)	0.35 (0.16–0.55)	p<0.007

**Table 4** Sensitivity, specificity, PPV, and NPV of PSQ using  $\text{oAHI} \geq 5$  events-h-1 for children 2–18 years of age

	Number of patients completed the PSQ (%)	Sensitivity % [95% CI]	Specificity % [95% CI]	PPV %	NPV %
All disease ( <i>n</i> : 588)	429 (72.9)	71.8	40.4	27.6	81.9
Chronic lung disease ( <i>n</i> : 90)	59 (65.5)	85.7	34.6	15	94.7
Genetic-metabolic syndrome ( <i>n</i> : 69)	47 (68.1)	63.6	32	45.1	50
Neuromuscular disease ( <i>n</i> : 76)	67 (88.1)	87.5	54.2	20.5	96.9
Endocrine diseases ( <i>n</i> : 49)	39 (79.5)	66.6	44.4	34.7	75
Cerebral palsy ( <i>n</i> : 68)	48 (70.5)	88.8	30.7	22.8	92.3
Healthy ( <i>n</i> : 69)	56 (81.1)	64.2	40.4	26.4	77.2
Other ( <i>n</i> : 62)	113 (67.6)	46.1	42.3	28.5	61.1

CI, confidence interval

**Table 5** Treatment modalities of the patients

	<i>n</i> (%)
Non-invasive ventilation (NIV)	190 (62.5)
Supplemental oxygen	40 (13.1)
Adenotonsillectomy	21 (7)
Continuation of invasive ventilation	18 (6)
Decannulation	19 (6.2)
Anti-inflammatory treatment	14 (4.6)
Weight loss	2 (0.6)

problems than children without the condition [12, 13]. Different screening questionnaires were used for cerebral palsy, and OSA was detected at a high rate [12, 14, 15]. In the study in which Garcia et al. used PSQ to see OSA, significantly higher values were found in the cerebral palsy group, similar to our study [16]. Another study stated that the PSQ could be used safely in identifying OSA in CP patients [17].

One of the factors contributing to the low PSQ's sensitivity–specificity in detecting OSA in our study may be the diversity of the patient groups. Since the reason for OSA in each patient group differed, the PSQ questions may need to be more specific in screening different disease groups. Like our study, Pabary et al. found that the PSQ was ineffective as a reliable screening instrument for OSA [6]. Another reason may be that most patients with OSA were in the metabolic-genetic syndrome group. While the PSQ is a validated questionnaire for OSA in screening healthy children [11], it did not show the same value in screening patients with underlying diseases. The study results of Cielo et al. in syndromic patients with craniofacial problems also support our study results [18].

Although screening questionnaires are low-cost and straightforward tools, they can not replace overnight PSG. The consensus is that children who display symptoms and signs suggestive of OSA should have their clinical evaluation include them since they are helpful, especially

in low-resource settings. According to the AAP Clinical Practice Guideline, the European Respiratory Society (ERS) Statement on OSA in children, and the AASM statement, the PSG is the most reliable way to diagnose OSA [2, 7, 10]. In the past few years, studies examining the accuracy of at-home sleep apnea tests in analyzing OSA and whether they can replace PSG have increased because of the restricted availability of PSG laboratories [19–21]. Cheung et al. compared polygraphy with pulse transit time and PSG in 45 patients and showed that polygraphy with pulse transit time was an alternative to PSG [19]. Oceja et al. stated that PG was a valid, cost-effective method like PSG and could be used reliably in treatment planning without PSG [20]. The AASM recommends portable monitoring instead of PSG for diagnosing adults with an elevated risk of moderate-to-severe OSA lacking comorbidities, but they do not recommend this for children [22]. On the other hand, ERS recommends using PG when PSG is unavailable [7].

Nearly more than 50% of the individuals in our study had OSA. Our higher rate of OSA compared to previous studies might be due to being a tertiary reference center and higher referral rates of patients with underlying medical conditions. The patient group with the most significant number of OSA in this study was the genetic-metabolic syndrome group. The literature has reported that the frequency of OSA increases in genetic and metabolic diseases with craniofacial defects, consistent with this study's results [18, 23, 24].  $\text{OAHl}$  and  $\text{AHI}$  were higher in patients with genetic-metabolic disease; 32.6% of this group consisted of Down syndrome (DS) patients. OSA is common in children with DS due to many predisposing factors such as macroglossia, midface hypoplasia, overweight/obesity, and hypotonia [25]. Our results were similar to previous studies.

CAI was considerably elevated in endocrine disease patients in our study. It could result from the high prevalence of PWS patients (63.4%) in our study group. It has been reported that, together with OSA and hypersomnolence, CSA is also frequently encountered in PWS [26, 27]. Also,

our patients with endocrine diseases were younger than the other patient groups (median 47 months), like Cohen et al.'s study, in which the frequency of central apnea was higher in the infant group [28].

Adenotonsillectomy (AT) is currently one of the primary treatments for moderate to serious OSA in adenotonsillar hypertrophy among kids [29]. Our AT and non-invasive ventilation (NIV) rates were 6.9% and 62.4%, respectively. In the current study, low rates of AT and high rates of NIV as treatment options could result from elevated rates of patients with neuromuscular, genetic-metabolic, and chronic lung diseases. Like our study, Oros et al. found that NIV was more common than adenotonsillectomy in 108 patients with neuromuscular and genetic disorders [30].

The most significant restrictions of our study were that it was carried out in a single center and that patients with underlying severe illnesses comprised most of the study individuals. Therefore, it would only be appropriate to generalize our results to some of the population. Additionally, differentiation between central and obstructive hypopneas was not made while scoring sleep studies during the study period, which may cause an overestimation of OSA.

In conclusion, SDB is associated with significant morbidity when underdiagnosed and untreated early; children with challenging, long-term illnesses such as neuromuscular and genetic diseases with SDB symptoms should be evaluated for SDB. Questionnaires for evaluating SDB are not sensitive and specific for children with chronic conditions, and the best method for diagnosing OSA is still PSG. However, some studies showed that PSQ was useful to monitor response to OSA treatment as it includes patient symptoms [31]. PSQ can diagnose patients and evaluate their response to treatment when combined with objective data, such as PSG.

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**Data availability** I confirm I have included a data availability statement in my main manuscript file.

The datasets generated during and analyzed during the current study are available from the corresponding author upon reasonable request.

## Declarations

**Ethics approval** Approval was obtained from the ethics committee of Marmara University. The study was performed by the ethical standards as laid down in the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

**Informed consent** Informed consent was obtained from all individual participants included in the study.

**Competing interests** The authors declare no competing interests.

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