

# **aPrevalence of sleep breathing disorders and their effects on dentofacial features among a sample of preadolescent and adolescent dental patients in Royal Medical Services.**

*Dr.Ahmad Al-Tarawneh, BDS \*Dr.Raghda Al-Shammout, DDS \*\*,Dr.Ra'ed Al-Dboush, BDS \*\*,Dr.Anwar Al-Rahamneh, DDS \*\*,Dr.Ayman Alelaimat, BDS\*\**

## **ABSTRACT**

**Background:** Sleep-Disordered Breathing (SDB) and Obstructive Sleep Apnea (OSA) are common in childhood and can have a range of adverse health effects. Diagnosis and treatment of these sleep disorders at an early age will help reduce their negative impact on children's dentofacial structures.

**Objective:** This study aims to assess the prevalence of SDB among preadolescent and adolescent patients, to investigate its effect on dentofacial features and to assess the risk factors associated with SDB.

**Methods:** A cross-sectional study of 150 patients of either gender who attended dental clinics of the Jordanian Royal Medical Services (JRMS) was carried out. Patients included in this study were aged 6-18 years old. A consent form and questionnaire were distributed to the patients' parents. An Arabic version of Pediatric Sleep Questionnaire (PSQ) was used in this study. Orthodontic routine clinical examinations were conducted for each patient to record their accompanying dentofacial features.

**Results:** 20.5 % (95% CI: 13.1-28.0) of the study sample were at risk of SDB. Males were more likely to suffer from this problem. SDB symptoms were primarily associated with a morphological feature dolichofacial, reduced maxillary width and a high vault. So these features may be considered as a good predictor for diagnosing and early treatment of SDB.

**Conclusion:** The PSQ is a reliable and practical screening tool that should be implemented in daily dental practice. Using this tool, children with a high risk of SDB can be identified and referred for follow-up testing and management in order to reduce the effects of the disorder.

**Keywords:** Sleep breathing disorders, Obstructive Sleep Apnea, Pediatric Sleep Questionnaire, dental.

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## **Introduction**

Diagnosis of pediatric SDB can reflect a broad spectrum of symptoms and conditions, ranging from snoring to upper airway resistance syndrome to OSA.<sup>1</sup> OSA is defined as a disorder of breathing during sleep characterized by prolonged partial upper airway obstruction and/or intermittent complete obstruction that disrupts normal ventilation and normal sleep patterns.<sup>2</sup>

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From the Departments of:

\*Consultant Orthodontist, private practice, Amman-Jordan

\*\*Department of Dentistry, Royal Medical Services, Amman-Jordan

Correspondence should be addressed to: Dr.Ra'ed Al-Dboush, E-mail: raedabbade@gmail.com

OSA is a common chronic disorder in children and adolescents and has a dramatic impact on their systemic health and development.<sup>3</sup> Signs of untreated sleep apnea in school-aged children may include bedwetting, poor school performance due to misdiagnosed attention deficit hyperactivity disorder (ADHD), aggressive behavior, or developmental delay.<sup>2</sup> Rare sequelae of untreated OSA include brain damage, seizures, coma, and cardiac complications.<sup>2,4,5</sup> These children also may experience impaired growth.<sup>2,4</sup>

The prevalence of childhood OSA is obscured by different diagnostic criteria that have been used in published studies. Epidemiologic data from 2008 indicates the prevalence of parent-reported 'always snoring' to be 1.5%-6%, the prevalence of parent-reported apneic events during sleep to be 0.2%-4%, and OSA diagnosed by varying criteria to be 1%-4%.<sup>6</sup>

Polysomnography (PSG), or overnight sleep studies, is the gold method to the diagnosis of Pediatric OSA and the apnea-hypopnea index (AHI), or the number of apneas and hypopnea per hours of total sleep time,<sup>7</sup> This procedure, however, is expensive, time-consuming and labor-intensive. Instead, one potential screening tool that has been validated and used in orthodontic offices is the Pediatric Sleep Questionnaire (PSQ). This questionnaire has a positive predictive value of 0.4 (i.e., 40% of patients with a positive PSQ score will be diagnosed with OSA) and a negative predictive value of 0.99 (i.e., only 1% of patients with a negative PSQ score will be diagnosed with OSA). The PSQ is a valuable first step in screening patients who come into the orthodontic office without a history of OSA.<sup>6</sup> This validated questionnaire was approved by the American Association of Orthodontics (AAO) and the European Respiratory Society to be used as a screening tool to identify cases of SDB and patients with a high risk of OSA.<sup>7,8</sup>

This pioneer study aimed to determine the prevalence of SDB symptoms among Jordanian children aged 6-18 years in JRMS hospitals based on a parental report, assess associated risk factors and evaluate the dentofacial features in patients with a high risk of SDB.

The specialty of dentofacial orthodontics isn't limited to dealing only with malocclusions but the management of SDB bears witness to this. There is increasing interest in the role of the orthodontist and pediatric dentist, both are responsible for screening for SDB and valuable part in the multidisciplinary management of SDB in both children and adults.<sup>6,9</sup>

## **Material and methods**

### **Population:**

This cross-sectional study was conducted at JRMS hospitals. Prior to the commencement of the study, ethical approval was obtained from the Human Research Ethics Committee at the JRMS under number 42/8/2019 and all research steps were carried in accordance with the ethical principles of the Helsinki Declaration. All patients' parents were informed about the aims and methods of this study and they provided written consent to participate.

The sample consisted of 150 patients of either gender who attended orthodontic and pediatric dental clinics in the Royal Rehabilitation Center, Queen Rania Hospital and Princess Haya Hospital in Jordan between August and October 2019. The patients were 6-18 years old. Exclusion criteria included patients who had already started their orthodontic treatment or those who had been diagnosed with a congenital anomaly or syndrome.

Patients were considered to be a cross-section of all layers of society, having different backgrounds, incomes and educational levels.

## **Questionnaire**

The PSQ was used to record the symptoms of SDB and to estimate the prevalence of parent-reported OSA. The PSQ was designed in 2000 by Chervin et al. and validated as a reliable parent-reported screening instrument for pediatric OSA (age range from 2 to 18 years old) and to predict sleep-related breathing disorders for use in clinical research.<sup>10</sup> The Sleep-Related Breathing Disorder (SRBD) scale of the PSQ is a 22 items survey that asks questions related to snoring and observed apnea, daytime sleepiness and inattentiveness, and other symptoms characteristic of childhood OSA. The presence of OSA can be predicted by the questionnaire with a sensitivity level of 81% and a specificity level of 87%. Based on the data of Chervin et al., the PSQ predicted OSA-related neurobehavioral morbidity as well as or better than polysomnography did.<sup>11</sup> In 2014, De Luca Canto et al. performed a systematic review to evaluate the diagnostic capabilities of various questionnaires and clinical examinations for pediatric SDB. They concluded that only the PSQ had enough diagnostic accuracy to warrant its use as a ‘screening method’ for pediatric SDB.<sup>12</sup> An Arabic version of the questionnaire was validated in the neighboring and Arabic-speaking Saudi Arabia. The original PSQ was translated into Arabic using the forward-backward translation method described by the World Health Organization (WHO) and the Arabic version was translated back into English by two bilingual dentists. The two versions were found to be consistent.<sup>13</sup>

The 22 items questionnaires were distributed to patients’ parents and they were given time and privacy to answer them. One of the authors was available to clarify any items that were unclear to the parents. To maximize returns, questionnaires were distributed, filled in and collected in one visit. Before commencing the research, a pilot study with ten patients was undertaken (not included in the final study) to test the clarity of the questionnaire and the phrases used. Routine orthodontic examinations were done as part of the regular visits of the patients. The dentofacial features that could be related to sleep disorders were extracted from the examination forms and reported.

## **Scoring SRBD Scale of PSQ**

The 22 items of the SRBD Scale were each answered using the following system: ‘Yes’ = 1, ‘No’ = 0, or ‘I don’t know’ = missing. The number of symptom-items answered positively (Yes) were divided by the number of items answered ‘Yes’ or ‘No’; the denominator therefore excluded items with missing responses and items answered as ‘I don’t know’. The result is a proportion that ranges from 0.0 to 1.0. Scores > 0.33 are considered positive and suggestive of high risk for pediatric SDB. Based on the PSQ scores, children with 33% or more positive responses (scores of eight or more) were considered at high risk of SDB, whereas children with fewer than 33% positive responses (scores less than eight) were considered at low risk of SDB.

## Statistical analysis

All data was collected, coded, and statistically analyzed using SPSS version 25 (IBM SPSS Statistics for Windows, Version 25.0; IBM, Armonk, NY, USA). Participants' characteristics and prevalence rates were reported using descriptive statistics. Characteristics of participants were described using frequency distribution tables for categorical variables and mean/standard deviation for continuous variables. Bivariate analyses were performed using the Pearson Chi-square ( $\chi^2$ ) Test to assess differences between the low- and high-risk groups regarding gender, snoring, difficulty in breathing, sleep apnea, mouth-breathing, daytime sleepiness/development, nocturnal enuresis (bedwetting), obesity, inattentive/hyperactive behavioral features, and orofacial features. Fisher's Exact Test was employed when applicable. Multivariate logistic regression was used to address the risk factors associated with the high-risk group. Odds Ratios (OR) and their 95% Confidence Intervals (CI) were reported. A p-value of less than .05 was considered statistically significant. The Hosmer–Lemeshow Goodness-of-Fit Test and the area under the Receiver Operator Characteristic (ROC) curve were used to assess the validity of the multivariate regression model.

## Results

One hundred and fifty patients attended three dental clinics in the different JRMS centers. Two of these centers are located in Amman, the capital of Jordan, and the 3<sup>rd</sup> in the northern city of Ajloun. 38 patients were excluded based on the exclusion criteria as they had already started their orthodontic treatment. Ultimately, 112 patients met the inclusion criteria stated previously and consent forms were collected and signed by parents. The included patients' parents were asked to complete the questionnaire. The response rate for the distributed questionnaires was 100%. This high rate was obtained due to the questionnaires being filled out and collected at the same time/in the same visit.

The study participants were 112: 60 were male (53.6%) and 52 were female (46.4%). The mean age of the patients was  $11.59 \pm 2.95$  years. The Kolmogorov-Smirnov Test with a Lilliefors Significance Correction was done to test the normality of the age variable.

In total, 20.5 % (95% CI: 13.1-28.0) of the children (23 patients out of the 112) in this study were at high risk of SDB (eight or more responses were 'Yes' based on the PSQ). Of the 23 patients who were categorized as high-risk, 19 were male and four were female. Regarding the SDB symptoms, snoring throughout sleep was reported in 15.2% of children, snoring loudly in 8%, sleep apnea in 3.6%, mouth-breathing in 30.4%, obesity in 9.8%, and bedwetting in 10.7%. Table 1 summarizes the distribution of participants with SDB symptoms (frequency and percentage).

Males were at a higher risk of having SDB than females ( $p = .003$ ). Sleep symptoms, daytime sleepiness and other SDB symptoms were strongly related to the high-risk group based on the PSQ. 21.7% of the high-risk group reported Temporomandibular Joint Disorders (TMD) symptoms ( $p = .004$ ) but there was no significant relationship between the age groups (6-9, 10-18 years) and the risk of SDB ( $p = .577$ ). Table 2 shows the bivariate analysis of the children's characteristics with regards to the high- and low-risk groups.

Multivariate logistic regression analyses to assess possible risk factors for OSA are presented in Table 3. Males presented a five times higher risk of developing SDB problems than females did (OR 5.4, C.I: 1.396-20.568,  $p = .014$ ). Obese children were nine times more likely to suffer from SDB or OSA (OR 8.704, C.I: 1.835-41.280,  $p = .006$ ). Children whose parent reported them to be suffering from nocturnal enuresis were nine times more likely to be in the high-risk group based on the PSQ (OR 8.502, C.I: 1.692-42.716,  $p = .009$ ).

## Discussion

SDB characterized by upper airway obstruction ranges from the cardinal symptom of primary snoring to a complete cessation of oxygen flow. The impact of this disease on patients, their families, and the healthcare system warrants increased attention and orthodontists can play an integral role.<sup>14</sup> One potential screening tool that has been validated and used in orthodontic offices is the PSQ. It is a user-friendly questionnaire designed specifically for pediatric patients. The PSQ is a valuable first step in screening patients who come into the orthodontic office without a history of OSA.

In this study, 20.5% of children were identified as high-risk for SDB, which is similar to the results reported in various other studies around the world. The results in other countries were as follows: Saudi Arabia 21%,<sup>13</sup> Netherlands 25%,<sup>15</sup> Chile 17.7%,<sup>16</sup> New Zealand 17.5%.<sup>17</sup> The results of this study were higher than that reported in the USA (7.3%)<sup>14</sup> and Belgium (4.1%),<sup>18</sup> while in Malaysian children the prevalence of the parental report of SDB symptoms was 14.9%.<sup>7</sup>

Habitual snoring is an alarming symptom of SDB. As reported by their parents, 6.3% of the patients suffered from this symptom, which is similar to the percentage reported in Turkey of 7%,<sup>21</sup> and Portugal of 8.8%,<sup>22</sup> but higher than that reported in Italy of 4.9%<sup>23</sup> and in Greece of 4.2%.<sup>24</sup> It is, however, lower than that reported in Hong Kong (10.9%),<sup>25</sup> India 11.4%,<sup>26</sup> Saudi Arabia 14.4%,<sup>13</sup> and Brazil 27.6%.<sup>27</sup>

Sleep apnea was reported in 3.6% of the total patients, which is higher than that witnessed in children in Hong Kong (1.5%)<sup>25</sup> and Brazil (0.8%).<sup>27</sup> Bedwetting was reported in 10.7% of patients, which is similar to that reported in the Indian study of 8.7%,<sup>26</sup> slightly higher than that reported in Hong Kong (5.1%),<sup>25</sup> while lower than that reported in Saudi Arabia (20.3%).<sup>13</sup> 30.4% of the children in the study were reported as mouth-breathers, as opposed to the 15.5% of Brazilian children<sup>27</sup> and 21% of Saudi children.<sup>13</sup>

Sleepiness during the day is a serious problem that could impact the academic performance of the student. 11.6% of parents who participated in this study reported that have been previously notified by their child's teacher about his/her sleepiness in the classroom. Our result is similar to that reported in the Netherlands (10%)<sup>15</sup> and Saudi Arabia (9.9%),<sup>13</sup> while higher than that reported in India 6.9%,<sup>26</sup> and Brazil 7.8%.<sup>27</sup>

There was a strong association in this study between gender and the risk of SDB with more male predilection ( $p = .003$ ). The proportion of males and females who were categorized as high-risk was 31.7% and 7.7% respectively. This result coincides with the results of studies done in Greece,<sup>24</sup> Chile,<sup>16</sup> New Zealand,<sup>17</sup> Saudi Arabia,<sup>13</sup> Malaysia,<sup>7</sup> Turkey,<sup>21</sup> Italy<sup>23</sup> and Hong Kong,<sup>25</sup> which reported that males were more at risk of SDB than females. This is in contrast to studies done in India,<sup>26</sup> the Netherlands,<sup>15</sup> and Brazil,<sup>27</sup> which found that gender had no significant effect on the prevalence of sleep disorders. One study done in Belgium reported an equal proportion of sleep disorders between female and male groups.<sup>18</sup>

Regarding the dentofacial features, the dolichofacial profile was found more in the high-risk group (47.8%), as opposed to 24.7% in the low-risk group with a statistically significant difference ( $p = .005$ ). Reduced

maxillary width and a high vault were found to be significantly correlated with the high-risk group ( $p = .01$ ,  $p = .014$ , respectively). 21.7% of the high-risk group were reported to have TMD symptoms and there was a strong association between the presence of TMD symptoms and being at high risk of SDB based on the PSQ score ( $p = .001$ ). This was similar to results reported by Smith et al. and Lavigne et al.<sup>28,29</sup>

The relationship between Non-Nutritive Sucking (NNS) habits such as digit sucking and SDB is debatable. While Guimaraes et al. found that this prevents SDB,<sup>30</sup> Huynh et al. and Dibiase et al. reported a statistically significant association between a history of thumb/finger sucking and heavy breathing at night.<sup>13,31</sup> In this study, there was no statistically significant association between NNS habits and the risk of SDB ( $p = .748$ ). This finding was similar to that reported by Altalib et al.<sup>32</sup>

Multivariate statistical analysis to assess factors associated with the presence of SDB showed that the following are significant predictors of SDB: being male (OR = 5.4, CI 1.4-20.6,  $p = .014$ ), being a mouth-breather (OR = 6.8, CI 2.1-21.9,  $p = .001$ ), suffering from bedwetting (OR = 8.5, 95% CI 1.7-42.7,  $p = .009$ ) and obesity (OR = 8.7, CI 1.8-41.3,  $p = .006$ ). Obesity was the most significant predictor for the presence of SDB and those who suffered from this issue were nearly nine times more likely to be assessed as high-risk for SDB based on the PSQ score.

## CONCLUSIONS

This study was conducted to determine the prevalence of SDB and OSA problems in preadolescents and adolescents in Jordan. 20.5 % (95% CI: 13.1-28.0) of the study sample were at risk of SDB. Males were more likely to suffer from these issues. Orofacial symptoms (being dolichofacial, having reduced maxillary width and a high vault) were more common in high-risk children. Mouth-breathing, bedwetting and obesity were significant predictors of SDB. Screening for the high risk of SDB in dental offices is practical and feasible. With a reliable screening tool like the PSQ, high-risk children can be identified and referred for follow-up testing and management to reduce the harmful effects of SDB on their dentofacial structures and quality of life.

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Table 1: Distribution of participants with SDB symptoms (frequency and percentage)

Subscale	Question	N (%)
Nocturnal symptoms / breathing problems	snore more than half the time	14(12.5)
	always snore	7(6.3)
	snore loudly	9(8)
	have ``heavy" or loud breathing	15(13.4)
	have trouble breathing, or struggle to breathe	10(8.9)
	stop breathing during the night	4(3.6)
	Tend to breathe through the mouth during the day	34(30.4)
	Have a dry mouth on waking up in the morning	32(28.6)
	Occasionally wet the bed	12(10.7)
Daytime sleepiness and development	wake up feeling unrefreshed in the morning	61(54.5)
	have a problem with sleepiness during the day	40(35.7)
	Has a teacher or other supervisor commented that your child appears sleepy during the day	13(11.6)
	Is it hard to wake your child up in the morning	42(37.5)
	Does your child wake up with headaches in the morning	14(12.5)
	Did your child stop growing at a normal rate at any time since birth	3(2.7)
	Is your child overweight	7(6.3)
Inattention/ hyperactivity	does not seem to listen when spoken to directly	20(17.9)
	has difficulty organizing task and activities	22(19.6)
	is easily distracted by extraneous stimuli	43(38.4)
	fidgets with hands or feet or squirms in Seat	20(17.9)
	is `on the go' or often acts as if `driven by a motor'	20(17.9)
	interrupts or intrudes on others (e.g. butts into conversations or games)	33(29.5)
Number of children at high risk of OSA ( 8 or more yes answers )		23(20.5)

Table 2: children characteristics classified based on the risk of OSA (Bivariate Analysis)

Domain	Variable	Response	All children N=112(%)	Low risk group N=89 (%)	High risk group N=23(%)	P value
Personal characteristic	Gender	Female	52(46.4%)	46(51.7)	4(17.4)	0.003*
		Male	60(53.6)	43(48.3)	19(82.6)	
	Age group	<10 years	29(25.9)	22(24.7)	7(30.4)	0.577
		>=10 year	89(79.5)	67(75.3)	16(69.6)	
Nocturnal problems	Snoring	Yes	17(15.2)	7(7.9)	10(43.5)	<.001*
		Usually snoring	14(12.5)	7(7.9)	7(30.4)	0.008†
		ways 'Hab snoring	7(6.3)	2(2.2)	5(21.7)	<.004†
	Breathing difficulty	Yes	18(16.1)	10(11.2)	8(34.8)	0.011*
	Sleep apnea	Yes	4(3.6)	1(1.1)	3(13.0)	0.027*
	Mouth breathing	Yes	34(30.4)	20(22.5)	14(60.9)	<0.001*
	Bed wetting	Yes	12(10.7)	6(6.7)	6(26.1)	0.016†
Daytime sleepiness and development	Wakeup un refreshed	Yes	61(54.5)	43(48.3)	18(78.3)	0.01*
	Day time sleepiness	Yes	40(35.7)	25(28.1)	15(65.2)	0.001*
	Class sleepiness	Yes	13(11.6)	4(4.5)	9(39.1)	<0.001*
	Hard to wake up	Yes	42(37.5)	28(31.5)	14(60.9)	0.009*
	Headache at morning	Yes	14(12.5)	8(9)	6(26.9)	0.014*
	Stop growing?	Yes	3(2.7)	0(0)	3(13.0)	0.009*
	Over weight	Yes	11(9.8)	4(4.5)	7(30.4)	.001**
ADHA symptoms	Not responding quickly when spoken to	Yes	20(17.9)	10(11.2)	10(43.5)	0.001*
	Difficulty on organizing things	Yes	22(19.6)	10(11.2)	12(52.2)	<0.001*
	Easily distracted	Yes	43(38.4)	26(29.2)	17(73.9)	<0.001*
	Seems restless when seated	Yes	16(14.3)	6(6.7)	10(43.5)	<0.001*
	Looks in hurry all the time	Yes	17(15.2)	7(7.9)	10(43.5)	<0.001*
	Interrupts others while they speak	Yes	33(29.5)	18(20.2)	15(65.2)	<0.001*
Orofacial features	Face morphology	Dolicofaci	33(29.5)	22(24.7)	11(47.8)	0.005*
	TMD symptoms	Yes	7(6.3)	2(2.2)	5(21.7)	0.004*

Variable		Odds ratio	Confidence intervals		P value
Gender	Male vs. Female	5.358	1.396	20.568	.014**
Mouth breathing	Mouth breather vs NOT mouth breathe	6.764	2.090	21.897	.001**
Bed wetting	Yes vs no	8.502	1.692	42.716	.009**
Weight	Obese vs Not	8.704	1.835	41.280	0.006**

Drofacial featur	Face morphology	Dolicofaci	33(29.5)	22(24.7)	11(47.8)	0.005*
	TMD symptoms	Yes	7(6.3)	2(2.2)	5(21.7)	0.004*
	non-nutritive sucking habits	Yes	17(15.2)	13(14.6)	4(17.4)	0.748
	Maxillary width	Narrow	38(33.9)	25(28.1)	13(56.5)	0.01*
	Palatal vault	High vault	39(34.8)	26(29.2)	13(56.5)	0.014*

\*Statistically significant (P<0.05) based on Chi-square test.

† Statistically significant (P<0.05) based on Fisher exact test.

Table 3: Sleep-disordered Breathing Risk (Multivariate Logistic Regression analysis\*)

\*Homser and Lemeshow test =0.904

\*\*Statistically significant (P<0.05)