Sleep disordered breathing in children seeking orthodontic care

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Introduction: Our objective was to evaluate the prevalence of high-risk factors for sleep disordered breathing (SDB) in an orthodontic population of children. SDB is a spectrum of breathing disorders ranging from primary snoring to obstructive sleep apnea. **Methods:** The sample included 303 healthy children between the ages of 9 and 17. High risk of SDB was assessed using the Pediatric Sleep Questionnaire, a validated instrument that consists of 22 questions, and high risk is defined as positive answers to 33% or more of the questions answered. Sixteen randomly selected patients repeated the questionnaire 1 month after the initial survey for reliability. **Results:** In this sample, high-risk status on the Pediatric Sleep Questionnaire was not associated with sex, age, or race. The percentage of patients who were screened as high risk was 7.3% (95% confidence interval, 4.7%-10.6%). **Conclusions:** The results of this study suggest that approximately 7% of adolescent orthodontic patients may be at a significant risk for some form of SDB. (Am J Orthod Dentofacial Orthop 2018;154:65-71)

Sleep is a tightly regulated and well-organized biologic process that affects our daily functioning and our physical and mental health. According to the American Sleep Association, there are different stages of sleep, primarily separated into rapid eye movement sleep and nonrapid eye movement, which is additionally divided into 3 stages.[1] Humans spend almost 50% of total sleep in the second stage of nonrapid eye movement and about 20% in rapid eye movement sleep. Deep and restorative sleep occurs in the third stage, whereas rapid eye movement sleep provides energy to the brain and supports daytime performance.[1,2]

According to the *Journal of Clinical Sleep Medicine*, children aged 6 to 12 years should sleep 9 to 12 hours, and teenagers aged 13 to 18 years should sleep 8 to 10 hours per night, for the body to fully recover and properly function. On average, nearly half of childhood is dedicated to sleep.[3] However, for high school students in the United States, only 25% have reported sleeping 8 hours or more per night.[4] The health effects of chronic sleep deficiency are immune suppression, cardiovascular disease, neurologic imbalances, compromised quality of life, and ultimately a shortened lifespan.[1,5,6]

Several factors can interfere with sound sleep, including sleep disordered breathing (SDB). SDB describes a group of disorders characterized by abnormal respiratory patterns such as apneas or hypopneas or insufficient ventilation during sleep. This complex group of disorders occurs during sleep and is described by extended periods of upper airway resistance (snoring) at 1 end of the spectrum and partial or complete airway obstruction at the other.[7]

Craniofacial morphology seems to be often correlated to SDB, particularly obstructive sleep apnea, which is related to neurogenic and anatomic factors. In the sagittal dimension, the most common findings in adults are an increased hyoid to mandibular plane angle and an increased length of the soft palate. A narrower posterior pharyngeal space has been found in adolescents who snore compared with those who do not.[8,9] Anatomically, the floor of the nose is the roof of the mouth. Transverse skeletal discrepancies as a result of maxillary constriction are associated with narrower lateral nasal walls, decreased nasal volume, and increased nasal airway resistance.[10] To compensate for the increased upper airway resistance, mouth breathing becomes the primary means of respiration.[11] Angle in

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1907 called mouth breathing the "most potent cause of malocclusion," and the literature has reports from as early as 1800 identifying airway obstructions as etiologies of malocclusions in both dental and medical fields.[12-14] It has also been reported that changes in the transverse dimension by using maxillary expanders can increase the airway volume in the nasal area.[15]

It is not often recognized that the clinical presentation of SDBs differs between children and adults. Adults become lethargic when sleepy, whereas hyperactivity is exacerbated in children when they do not have sufficient sleep. Thus, children with SDB may at times be misdiagnosed as having attention deficit disorder and subsequently medicated for the wrong condition.[16,17] Snoring, often thought to be a common symptom, is not generally reported to pediatricians; the literature shows that snoring during childhood is not normal.[16] Due to these variations in the signs and symptoms of SDB between adults and children, pediatric SDB may be underrecognized and underreported in clinical settings.

Definitive diagnosis of SDB, particularly the most severe form that is obstructive sleep apnea, is made by both clinical examination and polysomnography. However, due to limited accessibility, required training time, and high expense, polysomnography has barriers that limit its use.[18] Accordingly, several questionnaires have been developed to assess patients at high risk of SDB who require further evaluation. Of the questionnaires used in various studies, there is a widely reported range of validity and reliability. Furthermore, question selections based on individual studies have limited the universal application of most questionnaires.[19]

Chervin et al[20] sought to evaluate SDB and the related symptoms in children. A validated prospective Pediatric Sleep Questionnaire (PSQ) was developed for patients 2 to 18 years old to assess patient-reported or parent-reported snoring, apneas, sleepiness, and behavior disorders. This 22-item questionnaire evaluates 3 categories. Questions in the first category inquire about snoring, the second about sleepiness, and the third about behavior (particularly inattention and hyperactivity). Scoring on the PSQ ranges from 0 to 22 points. The last 6 questions on inattention and hyperactivity were adapted from the American Psychiatric Association's questionnaire for attention deficit hyperactivity disorder and linked with a response category well recognized in other population-based epidemiologic studies.[21]

It was estimated that about 80% of people with SDB remain undiagnosed and are unaware that they have the condition.[22] Dental specialists such as orthodontists generally see adolescent patients more often than do their medical colleagues.[23] This could contribute to not only educating the public, but also assessing the risk of SDB, alerting those with high risk to follow it up with a sleep physician for proper diagnosis and course of action. The prevalence of SDB in the United States in populations seeking orthodontic care has not been reported in the literature to our knowledge. The aim of this study was to evaluate the prevalence of positive or potential SDB in children in the orthodontic population. See Supplemental Materials for a short video presentation about this study.

MATERIAL AND METHODS

The Institutional Review Board at Case Western Reserve University in Cleveland, Ohio, approved this study. As part of the initial screening, in addition to a medical and dental history form, the PSQ was given when the patient was under 18 years old (Fig 1). The records of 303 children between the ages of 9 and 17 years who came to the orthodontic clinic between January 2014 and March 2016 were examined. This study was performed at 1 site only, and all data were evaluated and interpreted by 1 orthodontist and 1 statistician with a PhD degree in epidemiology.

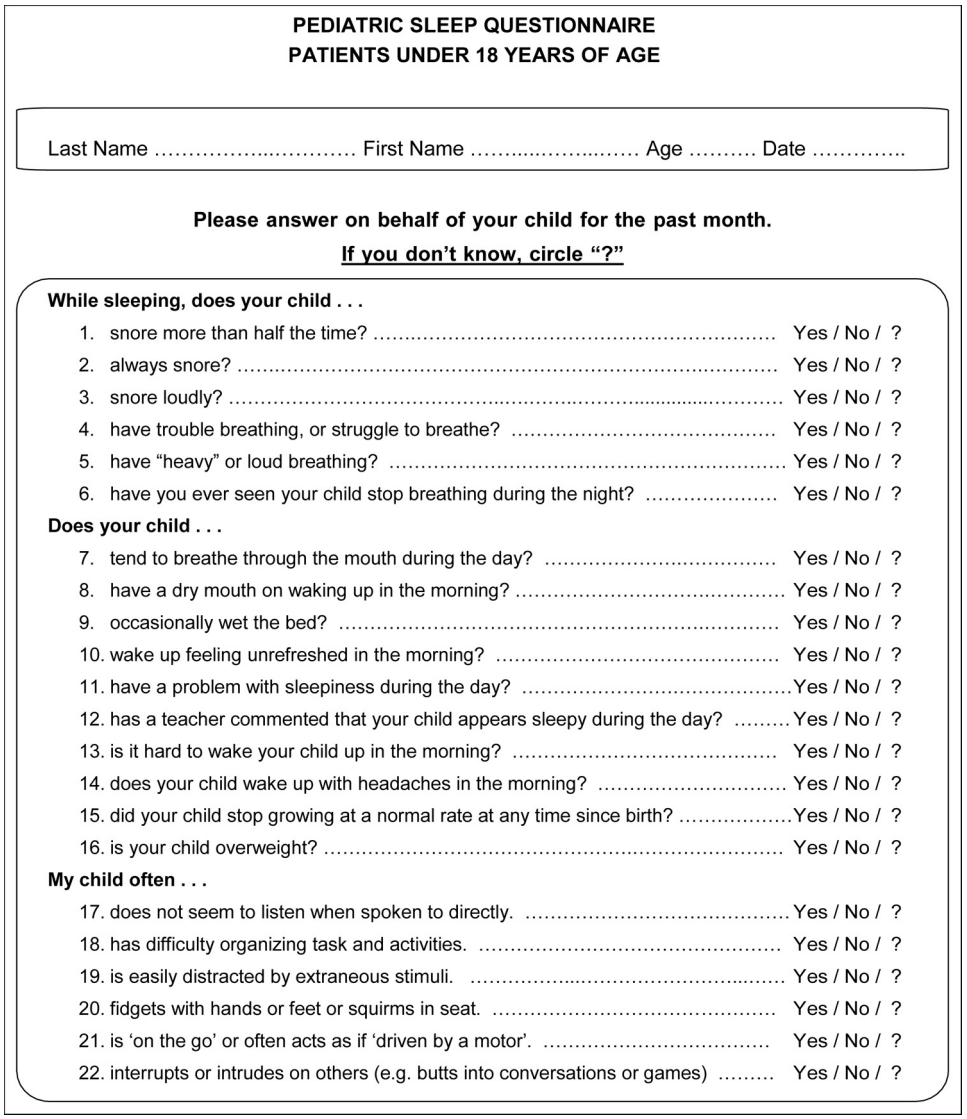
Inclusion criteria consisted of no previous orthodontic treatment, good general health, ability to read and write in English, and having completed the PSQ and health history forms. Patients with craniofacial anomalies, such as cleft lip and palate and syndromes, were excluded from this sample.

Patient age, sex, and ethnicity were collected on the PSQ. Racial and ethnic categories were defined according to the guidelines of the National Institutes of Health.[24] An additional category of "other" was listed as the seventh option choice for race. To evaluate the reliability of the PSQ in this study sample, 16 parents or legal guardians of the participants repeated the PSQ approximately 1 month after the initial completion.

On the PSQ, the response categories for all questions are "yes" or "no" or "don't know," indicated by "?". The cutoff value to identify patients at high risk for SDB is 0.33, meaning that 33% of the responses were "yes." A proportion of "yes" responses greater than or equal to 0.33 indicated a high risk of SDB; a lower percentage indicated a low risk. Missing and "don't know" responses were excluded from the denominator when estimating the percentage for risk status. When compared with polysomnographic data, the PSQ has previously demonstrated sensitivity of 0.85 and specificity of 0.87.[20]

Statistical analysis

All data were analyzed with the Statistical Package for the Social Sciences software (version 22.0; IBM, Armonk, NY). Descriptive statistics are presented (means



PEDIATRIC SLEEP QUESTIONNAIRE
PATIENTS UNDER 18 YEARS OF AGE

Last Name …… First Name …… Age …… Date ……

Please answer on behalf of your child for the past month.
If you don't know, circle "?"

While sleeping, does your child . . .

1. snore more than half the time? …… Yes / No / ?
2. always snore? …… Yes / No / ?
3. snore loudly? …… Yes / No / ?
4. have trouble breathing, or struggle to breathe? …… Yes / No / ?
5. have "heavy" or loud breathing? …… Yes / No / ?
6. have you ever seen your child stop breathing during the night? …… Yes / No / ?

Does your child . . .

7. tend to breathe through the mouth during the day? …… Yes / No / ?
8. have a dry mouth on waking up in the morning? …… Yes / No / ?
9. occasionally wet the bed? …… Yes / No / ?
10. wake up feeling unrefreshed in the morning? …… Yes / No / ?
11. have a problem with sleepiness during the day? …… Yes / No / ?
12. has a teacher commented that your child appears sleepy during the day? …… Yes / No / ?
13. is it hard to wake your child up in the morning? …… Yes / No / ?
14. does your child wake up with headaches in the morning? …… Yes / No / ?
15. did your child stop growing at a normal rate at any time since birth? …… Yes / No / ?
16. is your child overweight? …… Yes / No / ?

My child often . . .

17. does not seem to listen when spoken to directly. …… Yes / No / ?
18. has difficulty organizing task and activities. …… Yes / No / ?
19. is easily distracted by extraneous stimuli. …… Yes / No / ?
20. fidgets with hands or feet or squirms in seat. …… Yes / No / ?
21. is 'on the go' or often acts as if 'driven by a motor'. …… Yes / No / ?
22. interrupts or intrudes on others (e.g. butts into conversations or games) …… Yes / No / ?

Fig 1. Pediatric Sleep Questionnaire (PSQ).

and standard deviations or percentages) for quantitative and qualitative variables, respectively. PSQ scores were examined by sex and race categories with the Mann-Whitney test. Correlations of PSQ scores with age were evaluated with the Spearman rank correlation coefficient (rho). Cronbach's alpha for internal consistency was calculated from 36 responses for 3 subscales (snoring, sleepiness, and behavior as defined by Chervin et al[20]) and the total scale. The subscale and total scale scores from 2 administrations of the survey (test-retest) were compared with the Spearman rank correlation coefficient and Wilcoxon signed rank test. The test-retest portion had 16 participants.

RESULTS

Table 1 shows the study sample demographics. Twenty participants declined to identify their race. Sex was not associated with the race and ethnic categories.

The internal consistency of the responses on the 3 subscales (snoring, sleepiness, behavior) and the total scale were considered good (Cronbach's alpha, 0.719, 0.764, 0.754, and 0.730, respectively). The test-retest reliability was good for snoring (correlation coefficient rho, 0.730), but poor to moderate for sleepiness, behavior, and the total scale (correlation coefficient rho, 0.210, 0.445, and 0.230, respectively). However, no subscales or total score was significantly different

Table I. Characteristics of the study sample

		Frequency	*Valid percent (%)*
Age (y)	14.4 ± 2.02		
Sex (n)	Female	151	49.8
	Male	152	50.2
Race (n)	Asian	7	2.5
	Black	13	4.6
	Hispanic	17	6.0
	White	236	83.4
	Other	10	3.5
	Not reported	20	7.0
	Total	303	100.0

between the initial and follow-up PSQ (Wilcoxon signed rank test, all, $P > 0.257$).

The cutoff value to identify a patient as high risk for SDB was 0.33, or 33%, of "yes" responses. Of the 303 study participants, 22, or 7.3%, were identified as high-risk for SDB (95% confidence interval: 4.7%-10.6%). Overall, 144 (47.5%) of the questionnaires had no positive responses (Fig 2). Among the 22 high-risk patients, the mean proportion of "yes" responses was 42.6%, ranging from 33% to 77%. The proportion of boys and girls who were categorized as high risk (8.6% and 6.0%, respectively) was not significantly different ($P = 0.385$); SDB risk was also not associated with the race and ethnicity categories (Table II). Statistical testing of the cross-tabulation of risk category and race or ethnicity does not support a difference in the distribution of risk by race or ethnicity. However, these results should be viewed cautiously because of the small sample of participants in categories other than white. Age and PSQ scores were not correlated in this data set (Spearman rho, 0.098, $P = 0.09$).

DISCUSSION

SDB characterized by upper airway obstruction ranges from the cardinal symptom of primary snoring to complete cessation of oxygen flow. The impact of this disease on patients, their families, and the health care system warrants increased attention, and the orthodontist can play an integral role.

To date, there have been no studies in the United States with such a large cohort specifically evaluating the prevalence of SDB in an orthodontic population. Many authors, including Blunden et al,[16] have emphasized that primary caregivers underreport SDB. Underreporting is a contributing factor to underdiagnosis, and this problem is also caused by lack of awareness.

Previous studies have reported the prevalence of primary snoring to range from 3.2% to 12.1%.[25] In these studies, the sample populations primarily consisted of

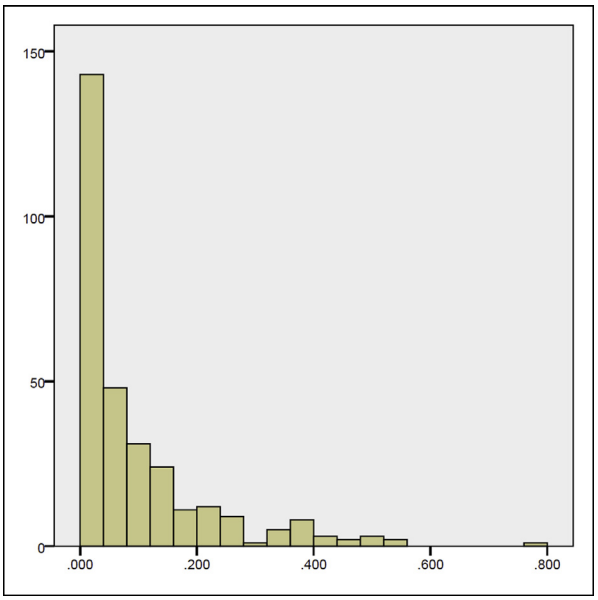


Fig 2. Frequency of the PSQ scored as a proportion of "yes" responses to the 22 questions, showing the results for the whole sample (n = 303). The x-axis represents the proportion of "yes" answers, and the y-axis represents the number of subjects in that percentage. For example, 144 subjects did not have a single "yes" in their responses, which is the longest bar on the left. Subjects to the right of 0.33 were considered at high risk for SDB.

Table II. Distribution of PSQ risk categories by sex and race

	High risk	*Low risk*	P *value*
Sex			0.385*
Female	9 (6.0%)	142 (94.0%)	
Male	13 (8.6%)	139 (91.4%)	
Race			0.794†
White	20 (8.5%)	216 (91.5%)	
Black	1 (7.7%)	12 (92.3%)	
Hispanic	1 (5.9%)	16 (94.1%)	
Asian	0	7 (100%)	
Other	0	10 (100%)	

*Chi-square test; †Fisher exact test.

preschool children. In our study, we estimated the prevalence of adolescents at high risk, which included primary snoring, for SDB at 7.3%.

According to information from the 2014 and 2015 reports from the American Association of Orthodontists Economics of Orthodontics survey, approximately 73% of patients in practices in the United States and Canada are under the age of 18 years.[26,27] American Association of Orthodontists members in the United States and Canada saw about 47 patients per day in their private

offices and worked 46 weeks annually. Since 7% to 10% of adolescent patients have some form of SDB, the average orthodontist in the United States sees 2 or 3 adolescents per day with some form of SDB, and this may be undiagnosed.[27,28] Furthermore, it was estimated that there were 5.4 million patients in active treatment in the United States and Canada in 2014; this translates to just less than 290,000 active child patients who could be at high risk for a sleep-related breathing disorder.[27]

Parents and guardians were compliant with completing the form which did not require provider time to administer it. In routine practice, a few minutes would be required to tally the score and then advise patients about positive screening results. Finally, neither our study nor previous reports addressed the issue of patient health literacy in completing the survey.

Although socioeconomic data were not collected in this study, there are reports in the literature on the direct relationship between socioeconomic status and health literacy and access to care.[29] Inclusion of these data could provide insight into the patients' chief concerns for seeking orthodontic treatment. On the department-issued medical and dental history form, there is a section of health concerns and a separate box for "other" whereby the patient can list other health concerns that are not already listed. For this study, an exact tally was not recorded for the number of patients who had adenoidectomy and tonsillectomy surgeries. However, for accurate health history reporting, this information was highlighted in the patient's chart. Our group did not want to introduce selection bias by preventing patients who had adenoidectomy and tonsillectomy surgery from being included in this study. If adenoidectomy and tonsillectomy surgery patients, who no longer have SDB, were excluded from this study, the prevalence may have been higher than the 7.3% that we are reporting.

Variations in study design and data reporting contributed to the differences of SDB prevalence in child patient populations. In the medical literature, prevalence estimates have ranged from 0.7% to 13%, since some have used solely oxygen saturation, whereas others were more comprehensive.[26] One study that looked at demographic data and medical risk factors for a large sample of children reported a lower SDB prevalence of 2.2%.[30] In a study evaluating craniofacial morphology and SDB using a modified version of the PSQ, breathing problems were reported to be as low as 1.8%, and snoring ranged from 2.9% to 10.9%. The more notable craniofacial findings from this study were that patients with dolichofacial facial patterns and steep mandibular plane angles were clinically and statistically more likely to have breathing problems.[31]

Our finding that 7.3% of the sample was at high risk for SDB was consistent with a 2008 meta-analysis that concluded that the prevalence of parent-reported snoring, the cardinal sign for SDB in children, was 7.45%.[25]

The PSQ was the primary screening tool used to evaluate SDB in our sample. Neither phenotypic presentation of the patient nor radiographic data were used in determining the prevalence. In their comprehensive review of the literature, Katyal et al[32] concluded that, although statistically notable, there was no clinically significant association between craniofacial morphology and pediatric SDB. Regardless, if this is the case, the PSQ stands alone as a validated screening tool for SDB risk, and it is often used in the medical literature to assess prevalence of SDB for specific populations.[33-37]

Self-reporting of the data may have limited reporting of the symptoms from parents or guardians, along with attitudes about why this information is necessary in the orthodontic clinic. This would lead to underreporting much like in other studies. Because of the retrospective and descriptive study design, the validity of these results cannot be objectively confirmed with polysomnography diagnosis. Patients surveyed were aged 9 to 17 years. Older patients in this group may not sleep in the same shared space as their parents, so lack of parent awareness of SDB symptoms may have influenced how questions were answered. The poorer reliability obtained in this study may have been influenced by the small sample size of repeated questionnaires and the possibility that, without our knowledge, older patients may have completed the responses at 1 time, and parents completed them at another time. Furthermore, small but frequent changes in responses among the low-risk group most likely contributed to lower reliability among all responses, without impacting the identification of risk status. Importantly, no high-risk subject in the initial PSQ changed risk status. Such factors were considered when the PSQ was independently assessed and should not significantly affect the quality of the results.[19]

We used a validated and calibrated screening tool (PSQ), which can be easily incorporated into a busy orthodontic office. It is a user-friendly questionnaire designed specifically for a pediatric population. Orthodontists are uniquely positioned to screen patients who may be at high risk for SDB but unaware of the condition. With a proper clinical examination and the use of screening questionnaires such as the PSQ, orthodontists can provide a more comprehensive health care service by potentially identifying and properly referring a high-risk patient to a pediatric sleep specialist, a pediatrician, or an otolaryngologist for a proper diagnostic workup and appropriate course of action. All 22 patients who were identified as high risk were referred to pediatric

sleep medicine for further evaluation. Outcome assessment was highlighted in the patient's health history.

CONCLUSIONS

The results of this study suggest that approximately 7% to 10% of adolescent orthodontic patients may be at significant risk for some form of SDB. Screening for the high risk of SDB in orthodontic settings is practical and feasible; with the proper screening tool, high-risk children can be identified and referred for further follow-up testing.

SUPPLEMENTARY DATA

Supplementary data related to this article can be found online at https://doi.org/10.1016/j.ajodo.2017.11.027.

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