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Correlation between intraoral markers and the risk of obstructive sleep apnea in children

A thesis submitted in partial fulfillment of the requirements for the degree of Master of Science
in Dentistry at Virginia Commonwealth University.

By

Linda K. Powers, DDS

The University of Maryland, College Park, 2015

The University of Maryland School of Dentistry, 2019

Thesis advisor: Eser Tüfekçi, DDS, MS, PhD, MSHA

Department of Orthodontics

Virginia Commonwealth University

Richmond, Virginia

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Abstract

CORRELATION BETWEEN INTRAORAL MARKERS AND THE RISK OF OBSTRUCTIVE SLEEP APNEA IN CHILDREN

By: Linda K Powers, DDS

A thesis submitted in partial fulfillment of the requirements for the degree of Master of Science in Dentistry at Virginia Commonwealth University.

Virginia Commonwealth University, 2023

Thesis Advisor: Eser Tüfekçi, DDS, MS, PhD, MSHA

Department of Orthodontics

Specific aims: The purpose of this study was to explore the association between various intraoral markers and the risk of Obstructive Sleep Apnea (OSA) in children. Specifically, nine intraoral clinical characteristics were analyzed: ankyloglossia, palatal vault height, presence and level of dental wear, tonsillar grade, Friedman's classification, skeletal and dental classification, and posterior crossbite. The correlation between these features and the Pediatric Sleep Questionnaire (PSQ) and Pediatric Symptoms Checklist (PSC) scores were assessed.

Background: Pediatric OSA is a condition in which there is the total or partial closure of the airway while a child is sleeping, which can have profound detrimental impacts on the child's development yet goes largely undiagnosed. While previous research on pediatric OSA focuses primarily on how OSA affects the oral cavity and OSA risk concerning extraoral markers, to date, increased sleep apnea risk in children has not been investigated concerning intraoral features.

Methods: Orthodontic patients aged 8-17 were recruited at the VCU Graduate Clinic. Intraoral measurements were collected from patients (n=100) by treating doctors. At this time, the PSQ and PSC screening surveys were also completed by the parents/legal guardians of the subjects. Analyses were performed to determine the association between the intraoral markers and the risk of pediatric OSA.

Results:

Eighteen percent of the subjects demonstrated a high risk for sleep-disordered breathing based on the PSQ survey and 10% for emotional or behavioral problems based on the PSC survey. There was a strong association between PSQ and PSC for high risk for OSA (p-value<0.0001). None of the intraoral markers demonstrated statistically significant associations with sleep-disordered breathing as measured by the PSQ. However, subjects deemed high risk of OSA based on the PSQ survey were more likely to have a higher tonsillar grade, a higher Friedman Classification, and a more shallow palatal vault.

Conclusions: In this study, 18% of subjects were concluded to be at high risk for OSA, as determined by the validated PSQ. While none of the intraoral markers demonstrated statistically significant associations with sleep-disordered breathing as measured by the PSQ, some variables showed trends. Future studies with a larger sample size are needed to investigate further a possible association between the clinical features and OSA.

Introduction

While an orthodontist's role is primarily to correct malpositioned teeth, as healthcare providers, they also have the potential to serve their patients in a greater capacity. Obstructive sleep apnea (OSA) has been a topic of increasing interest within orthodontics. In 2017 the American Association of Orthodontics formed a task force consisting of dental and medical sleep medicine experts to offer guidance on the role of the specialty in the management of OSA.¹ Based on the information compiled by a panel, it was concluded that orthodontic treatment has no proven etiology in developing OSA, and some treatment modalities may help manage the condition.¹

The task force also recommended that a sleep medicine make the diagnosis; however, orthodontists should become familiar with the signs and symptoms of the disorder. This way, they can be crucial in screening patients for sleep-disordered breathing (SDB).¹ Moreover, an understanding of SDB would help orthodontists participate in the multidisciplinary care for patients diagnosed with OSA. As children and adolescents comprise a large proportion of the orthodontic patient base, screening for pediatric OSA is an area where orthodontists have the potential to play a significant role in this public health issue.

SDB, a pathophysiologic continuum characterized by abnormal respiratory patterns, is common in the pediatric population, with a prevalence of 3 - 12%.² The symptoms can range

from snoring to obstructive sleep apnea. When a child experiences OSA, there is total or partial airway closure while sleeping. The pathophysiology is a complex interplay of anatomic variations that may predispose a child to airway obstruction and loss of neuromuscular tone. While many children exhibit intermittent snoring, according to the American Academy of Pediatrics,³ 1.2 to 5.7% of children are affected by pediatric OSA. Furthermore, it is thought that the prevalence of pediatric OSA is underestimated due to multiple factors, and if not treated, it may have important implications for the child's health.³

Pediatric OSA usually disrupts a restful night's sleep, resulting in daytime sleepiness, irritability, and general behavioral issues. OSA may ultimately impair the child's growth, cognitive development, and emotional stability in the long term.² These signs often go unnoticed or are attributed to another condition, such as attention deficit hyperactivity disorder (ADHD), because of an overlap between the symptoms of ADHD and OSA.⁴ These symptoms include hyperactivity, aggression, and irritability. Also, the manifestation of pediatric OSA is not uniform, and there is wide variability in its clinical presentation. For example, it can present as either fatigue or hyperactivity on the opposite side of the spectrum. Parents may also not recognize signs of OSA as an issue, with 80% of parents of symptomatic habitual snorers not reporting it to the pediatrician.⁵ These factors make screening for pediatric OSA difficult. When left untreated, poorly controlled pediatric OSA can result in serious complications such as cor pulmonale, pulmonary hypertension, and other chronic diseases. In addition, behavioral and psychosocial problems may be encountered, such as poor school performance.⁶

There is a higher prevalence of sleep-disordered breathing risk in orthodontic patients than in a healthy pediatric population. According to a recent study by Rohra et al.,⁷ an estimated 7.3% of adolescent orthodontic patients may be at significant risk for sleep-disordered breathing.

Using the screening tool used by Rohra et al.⁷ in a different orthodontic patient population, another study found the prevalence to be even higher, at 10.8%.⁸

In a 2022 study by Choong et al.,⁹ an astounding 30% of children seeking orthodontic care were reported to be at increased risk for OSA. Over the years, there has been a continuous increase in the prevalence of OSA. One explanation for this trend is that the cases were misdiagnosed mainly or undiagnosed in earlier studies. Nevertheless, there is still a movement towards more children experiencing the effects of disordered breathing, perhaps due to environmental factors such as diet and allergens causing obesity and allergic rhinitis.¹⁰

The current gold standard for diagnosing pediatric OSA is polysomnography (PSG), which generally involves an in-center overnight sleep study and analysis by a sleep medicine physician.¹ However, there are limited reliable sleep laboratories for children, leading to excessive waiting times to be scheduled for a study and, in general, difficulty in accessing care. There is also a substantial cost associated with such studies.²

One method of screening that has been validated is the Pediatric Sleep Questionnaire (PSQ). PSQ is a 22-question survey that analyzes snoring, daytime sleepiness, and abnormal behavior. The PSQ has a high sensitivity of 85% and specificity of 87% relative to PSG data for children ages 2-18. A 2014 systematic review and meta-analysis of SDB screening questionnaires demonstrated that only the PSQ survey had the diagnostic accuracy to be used as a screening tool for OSA in pediatric patients.¹¹ Although the PSQ is not deemed an acceptable replacement for PSG in the diagnosis of pediatric OSA, it is still a valuable tool for identifying children at risk for OSA.¹²

Since signs of untreated OSA include behavioral disturbances, another validated method of screening in children is the Pediatric Symptom Checklist (PSC), which eliminates the need for parents to know details of their child's sleeping behavior but focuses on daytime behaviors instead.¹³ This questionnaire can supplement the Pediatric Sleep Questionnaire to address the behavior components of OSA but is not intended as a tool to diagnose sleep-disordered breathing. While these tools are valuable, there is room for improvement in their positive predictive value. Furthermore, lengthy surveys are not practical in a busy orthodontic practice.

Research on children diagnosed with OSA shows the condition is associated with various oral and craniofacial manifestations. Pirilä-Parkkinen et al.¹⁴ examined the effects of sleep-breathing disorders on developing dental arches in a cross-sectional study. The authors noted that children diagnosed with OSA had a significantly increased overjet, a reduced overbite, and narrower upper and shorter lower dental arches when compared with age and gender-matched controls.¹⁴

Similarly, Flores Mir et al.¹⁵ reported that patients with a hyperdivergent growth pattern and a Class II malocclusion have a craniofacial morphology that resembles the features of pediatric OSA patients. In another study by Löfstrand-Tideström et al.,¹⁶ four-year-old children diagnosed with nocturnal breathing obstruction had a larger anterior facial height, a narrower maxilla, a deeper palatal height, a shorter lower dental arch, and a higher prevalence of lateral crossbite compared to asymptomatic children.¹⁶

Other possible dental variables related to pediatric OSA include occlusal wear, frenulum height, and tonsil size. Hosoya et al.¹⁷ found that individuals with OSA have a high risk of sleep bruxism in the general population. Another study reported that 36% of children with severe OSA have both snoring and sleep bruxism.¹⁸ In a retrospective study, Huang et al.¹⁹ noted that children

with untreated short frenulum developed abnormal tongue function early in life with a secondary impact on orofacial growth and sleep-disordered breathing.¹⁹

The literature has also well-established the relationship between tonsil size and pediatric OSA. Tonsillar size grading can be accomplished easily by observing where the tonsils are in relation to the tonsillar pillars.²⁰ Adenotonsillar hypertrophy, which can cause narrowing of the oropharynx leading to OSA, is reported to be the main factor contributing to breathing disorders in the pediatric population.² Li et al.²¹ measured tonsil size in 35 consecutive patients referred to a pediatric chest clinic because of suspected OSA. The authors suggested a positive correlation between the tonsil size and the severity of obstructive sleep apnea.^{20,21}

Another parameter that can affect airway patency is tongue position. The Friedman tongue position staging system has been shown to correlate with OSA severity in adults. However, similar results have not been demonstrated in children but are worth further investigation.²²

In the literature, most studies examined associations between extraoral factors such as obesity and neuromuscular diseases with OSA. The effects of OSA on the oral cavity have also been investigated. However, the intraoral characteristics such as retrognathia and deep palate, although considered predisposing factors for sleep disorders, have not been shown to have a strong relationship with OSA.¹ Nevertheless, based on a 2019 literature review, Luzzi et al.²³ concluded a strong association between craniofacial markers and pediatric OSA. The authors emphasized the role of dental professionals in recognizing possible pediatric OSA.²³

In 2022, a survey study by Berggren et al.²⁴ evaluated dental practitioners' perceptions of their ability to identify patients at risk for OSA. The results indicated that dentists and oral

hygienists lacked confidence in reliably recognizing the intraoral characteristics of OSA. Furthermore, they expressed a need for an available index or tool to achieve this task.²⁴

In light of conflicting literature on the association of intraoral measurements to OSA, there is a need to investigate if specific clinical characteristics could predict sleep apnea risk in children. Therefore, the purpose of this study was to determine if an association was present between the nine intraoral features and PSQ and PSC scores. If associations between intraoral markers and an increased risk of sleep apnea in children can be successfully identified, a screening tool could be developed. The availability of a reference guide could aid dental professionals in identifying patients at risk for sleep disorders during routine intraoral examinations.

Methods

This observational study was approved by the Institutional Review Board at Virginia Commonwealth University (VCU). The study sample included children seeking orthodontic treatment at the VCU graduate clinic. The inclusion criteria were that the subjects had to be 8-17 years old, and the patient and guardian had to be proficient in English. Patients with systemic diseases, craniofacial syndromes, and facial deformities were excluded from the study. During routine initial records appointments, treating doctors asked patients who met the inclusion criteria if they were interested in participating in the study. A minimum of 90 subjects was determined based on the rule of thumb of having ten individuals per predictor. A total of one hundred healthy, English-speaking subjects were recruited. Each patient was assigned a case number using a code such as Case 001. No individual identifying information was recorded for the analyses.

Once the informed consent was obtained, the parent was asked to complete the PSQ and PSC forms (Figures 3 and 4). The PSQ and PSC scores were calculated based on each screening instrument's guidelines. A PSQ score of over 33% positive answers and a PSC score of 28 or higher indicated a high risk for sleep-disordered breathing and behavioral health problems, respectively.

The child's orthodontic appointment proceeded as scheduled, which included a clinical examination, intraoral and extraoral photos, radiographs, and intraoral scanning of the teeth. In

addition to these routine procedures, a 2-3 minutes intraoral examination for the anatomical and dental characteristics was completed (Figure 1). These intraoral markers were selected based on previous literature suggesting an association between specific craniofacial features and the risk for sleep apnea.

The additional experimental intraoral screening consisted of recording data regarding the following nine items: the location and amount of dental wear, ankyloglossia, tonsillar grade, Friedman's Classification indicating tongue position, the relationship of the maxilla relative to the mandible (skeletal classification), Angle's classification of the malocclusion, palatal vault height, and transverse relationship for posterior crossbite. All clinicians were calibrated regarding how to measure each intraoral screening marker before data collection. Visual guides for measuring the intraoral variables were available at the initial appointment to guide the accurate data acquisition (Figure 2).

| | |
|--|---|
| <p>DATA COLLECTION FORM</p> <p>Age: _____</p> <p>Ankyloglossia (see A5)</p> <ul style="list-style-type: none"> <input type="radio"/> Normal (>16 mm) <input type="radio"/> I (12-16 mm) <input type="radio"/> II (8-11 mm) <input type="radio"/> III (3-7 mm) <input type="radio"/> IV (<3 mm) <p>Tonsillar Grade (see A1)</p> <ul style="list-style-type: none"> <input type="radio"/> 0 (not present) <input type="radio"/> 1 <input type="radio"/> 2 <input type="radio"/> 3 <input type="radio"/> 4 <p>Friedman's Classification (see A2)</p> <ul style="list-style-type: none"> <input type="radio"/> I <input type="radio"/> II <input type="radio"/> III <input type="radio"/> IV <p>Palatal Vault Height (see A3)</p> <ul style="list-style-type: none"> <input type="radio"/> Low <input type="radio"/> Medium <input type="radio"/> High | <p>Tooth Wear (see A4)</p> <ul style="list-style-type: none"> <input type="radio"/> Mild <input type="radio"/> Moderate <input type="radio"/> Severe <p>Spread of Tooth Wear</p> <ul style="list-style-type: none"> <input type="radio"/> Incisal <input type="radio"/> Posterior <input type="radio"/> Generalized <p>Molar Classification: (If first permanent molar is not present, use first primary molar)</p> <ul style="list-style-type: none"> <input type="radio"/> I <input type="radio"/> II <input type="radio"/> III <p>Skeletal Classification</p> <ul style="list-style-type: none"> <input type="radio"/> I <input type="radio"/> II <input type="radio"/> III <p>Posterior Crossbite</p> <ul style="list-style-type: none"> <input type="radio"/> None <input type="radio"/> Unilateral <input type="radio"/> Bilateral |
|--|---|

Figure 1: Data Collection Form

DATA COLLECTION FORM

Appendix

I. Tonsillar Grade



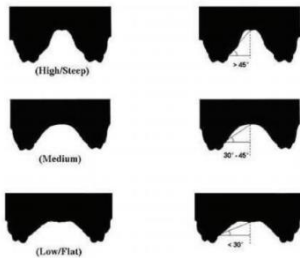
<https://sleepmedicineboardreview.wordpress.com/2011/10/25/tonsil-size-scoring/>

II. Friedman's Classification



<https://dimensionsofdentalhygiene.com/article/sleep-apnea-screening-in-the-dental-office/>

III. Palatal Vault Height



<https://www.ncbi.nlm.nih.gov/pubmed/18402092>

IV. Tooth Wear Score

| Score | Surface | Criteria |
|---------------|-----------------|---|
| 0 No wear | B/L/O/I C | No loss of enamel surface characteristics. No loss of contour. |
| 1 Mild | B/L/O/I C | Loss of enamel surface characteristics. Minimal loss of contour. |
| 2 Moderate | B/L/O I C | Loss of enamel exposing dentine for less than one third of surface. Loss of enamel just exposing dentine. Defect less than 1 mm deep. |
| 3 Moderate | B/L/O I C | Loss of enamel exposing dentine for more than one third of surface. Loss of enamel and substantial loss of dentine. Defect less than 1-2 mm deep. |
| 4 Severe | B/L/O I C | Complete enamel loss - pulp exposure - secondary dentin exposure. Pulp exposure or exposure of secondary dentine. Defect more than 2mm deep - pulp exposure - secondary dentine exposure. |

B: buccal; L: lingual; O: occlusal; I: incisal; C: cervical.

<https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3908810/>

V. Kotlow's Classification Technique: measure distance from tip of tongue to frenulum attachment



Figure 2: Data Collection Supplemental Information

Pediatric Sleep Questionnaire: Sleep-Disordered Breathing Subscale

070129

Child's Name: _____
 Person completing form: _____

Study ID #: _____
 Date: ____/____/____

Please answer these questions regarding the behavior of your child during sleep and wakefulness. The questions apply to how your child acts in general during the past month, not necessarily during the past few days since these may not have been typical if your child has not been well. You should circle the correct response or *print* your answers neatly in the space provided. A "Y" means "yes," "N" means "no," and "DK" means "don't know."

| | | | | |
|---|---|---|----|-----|
| 1. WHILE SLEEPING, DOES YOUR CHILD: | | | | |
| Snore more than half the time?..... | Y | N | DK | A2 |
| Always snore? | Y | N | DK | A3 |
| Snore loudly? | Y | N | DK | A4 |
| Have "heavy" or loud breathing? | Y | N | DK | A5 |
| Have trouble breathing, or struggle to breathe? | Y | N | DK | A6 |
| 2. HAVE YOU EVER SEEN YOUR CHILD STOP BREATHING DURING THE NIGHT? | | | | A7 |
| 3. DOES YOUR CHILD: | | | | |
| Tend to breathe through the mouth during the day?..... | Y | N | DK | A24 |
| Have a dry mouth on waking up in the morning? | Y | N | DK | A25 |
| Occasionally wet the bed? | Y | N | DK | A32 |
| 4. DOES YOUR CHILD: | | | | |
| Wake up feeling unrefreshed in the morning? | Y | N | DK | B1 |
| Have a problem with sleepiness during the day? | Y | N | DK | B2 |
| 5. HAS A TEACHER OR OTHER SUPERVISOR COMMENTED THAT YOUR CHILD APPEARS SLEEPY DURING THE DAY? | | | | B4 |
| 6. IS IT HARD TO WAKE YOUR CHILD UP IN THE MORNING? | | | | B6 |
| 7. DOES YOUR CHILD WAKE UP WITH HEADACHES IN THE MORNING?..... | | | | B7 |
| 8. DID YOUR CHILD STOP GROWING AT A NORMAL RATE AT ANY TIME SINCE BIRTH? | | | | B9 |
| 9. IS YOUR CHILD OVERWEIGHT? | | | | B22 |
| 10. THIS CHILD OFTEN: | | | | |
| Does not seem to listen when spoken to directly. | Y | N | DK | C3 |
| Has difficulty organizing tasks and activities. | Y | N | DK | C5 |
| Is easily distracted by extraneous stimuli. | Y | N | DK | C8 |
| Fidgets with hands or feet or squirms in seat. | Y | N | DK | C10 |
| Is "on the go" or often acts as if "driven by a motor". | Y | N | DK | C14 |
| Interrupts or intrudes on others (eg, butts into conversations or games). | Y | N | DK | C18 |

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Figure 3: Pediatric Sleep Questionnaire

Child's Name _____
 Today's Date _____
 Date of Birth _____

Record Number _____
 Filled out by _____

Pediatric Symptom Checklist

Emotional and physical health go together in children. Because parents are often the first to notice a problem with their child's behavior, emotions or learning, you may help your child get the best care possible by answering these questions. Please mark under the heading that best fits your child.

| | | Never (0) | Sometimes (1) | Often (2) |
|-----|---|--------------|------------------|--------------|
| 1. | Complains of aches/pains | 1 | _____ | _____ |
| 2. | Spends more time alone | 2 | _____ | _____ |
| 3. | Tires easily, has little energy | 3 | _____ | _____ |
| 4. | Fidgety, unable to sit still | 4 | _____ | _____ |
| 5. | Has trouble with a teacher | 5 | _____ | _____ |
| 6. | Less interested in school | 6 | _____ | _____ |
| 7. | Acts as if driven by a motor | 7 | _____ | _____ |
| 8. | Daydreams too much | 8 | _____ | _____ |
| 9. | Distracted easily | 9 | _____ | _____ |
| 10. | Is afraid of new situations | 10 | _____ | _____ |
| 11. | Feels sad, unhappy | 11 | _____ | _____ |
| 12. | Is irritable, angry | 12 | _____ | _____ |
| 13. | Feels hopeless | 13 | _____ | _____ |
| 14. | Has trouble concentrating | 14 | _____ | _____ |
| 15. | Less interest in friends | 15 | _____ | _____ |
| 16. | Fights with others | 16 | _____ | _____ |
| 17. | Absent from school | 17 | _____ | _____ |
| 18. | School grades dropping | 18 | _____ | _____ |
| 19. | Is down on him or herself | 19 | _____ | _____ |
| 20. | Visits doctor with doctor finding nothing wrong | 20 | _____ | _____ |
| 21. | Has trouble sleeping | 21 | _____ | _____ |
| 22. | Worries a lot | 22 | _____ | _____ |
| 23. | Wants to be with you more than before | 23 | _____ | _____ |
| 24. | Feels he or she is bad | 24 | _____ | _____ |
| 25. | Takes unnecessary risks | 25 | _____ | _____ |
| 26. | Gets hurt frequently | 26 | _____ | _____ |
| 27. | Seems to be having less fun | 27 | _____ | _____ |
| 28. | Acts younger than children his or her age | 28 | _____ | _____ |
| 29. | Does not listen to rules | 29 | _____ | _____ |
| 30. | Does not show feelings | 30 | _____ | _____ |
| 31. | Does not understand other people's feelings | 31 | _____ | _____ |
| 32. | Teases others | 32 | _____ | _____ |
| 33. | Blames others for his or her troubles | 33 | _____ | _____ |
| 34. | Takes things that do not belong to him or her | 34 | _____ | _____ |
| 35. | Refuses to share | 35 | _____ | _____ |

Total score _____

Does your child have any emotional or behavioral problems for which she/he needs help? N Y
 Are there any services that you would like your child to receive for these problems? N Y

If yes, what services? _____

Figure 4: Pediatric Symptom Checklist

Statistical Methods

Data from the PSQ and PSC questionnaires and the intraoral marker evaluation forms were entered into a database on Research Electronic Data Capture (REDCap) tool hosted at Virginia Commonwealth University.

Responses from PSQ and PSC screening tools were summarized. Associations between the intraoral measures and the risk for sleep-disordered breathing and behavioral health problems were assessed using Fisher's Exact test for the PSQ and PSC scores, respectively. The significance level was set at 0.05. SAS EG. v.8.2 (SAS Institute, Cary, NC) was used for all analyses.

Results

One hundred patients were recruited for the study. These subjects were screened for the nine intraoral markers during their initial orthodontic examination. The guardians of the participants also filled out the Pediatric Sleep Questionnaire and Pediatric Symptoms Checklist. According to the PSQ scores, eighteen subjects (18%) were identified as at high risk for sleep-disordered breathing. Similarly, the PSC grading showed ten individuals (10%) with an increased risk for emotional or behavioral problems.

The summary statistics of the intraoral markers are presented in Table 1. Most patients had a normal tongue structure (87%). Ankyloglossia, a condition restricting the tongue's motion of range, was detected in only 13% of patients. Similarly, most patients had an average tonsillar grade (0 or 1), but only 38% of the subjects had enlarged tonsils (2 and 4). Eighty-three percent of the patients exhibited an increased Friedman stage (III and IV), a condition suggestive of upper airway obstruction. As for the palatal anatomy, 75% of the subjects had a typical palatal vault height (medium). Regarding tooth wear, 92% of the individuals had signs of mild tooth wear. Finally, 61% of patients had a Class I molar relationship, while 66% of the subjects had a Class I skeletal classification. Most of the participants had no posterior crossbite (89%).

A strong association existed between the results of the PSQ and PSC screening tools (p-value<0.0001). Eighty percent of those determined by PSQ at high risk for sleep-disordered sleeping were also found at increased risk for behavioral health problems by the PSC tool. Only 11% of those determined by PSC not having behavioral health problems were found at high risk on the PSQ (Table 2).

None of the intraoral markers demonstrated statistically significant associations with sleep-disordered breathing as measured by the PSQ. However, tonsillar grade, palatal vault height, and Friedman Classification had a trend suggesting a possible correlation (Figures 5-7). Only five of the 18 patients at high risk by the validated PSQ screening tool had a tonsillar grade of 3. Although identified as at risk for pediatric OSA, 13 of these individuals had a tonsillar grade of 0/1 and 2 (p-value=0.2468). Fifty percent of subjects with a low palatal vault height had a high risk of SDB based on the PSQ, compared to 19% with an average vault height and 10% with a high vault height (p-value=0.1533). Participants with a Friedman Classification of III and IV, indicating upper airway obstruction, demonstrated a higher risk for SDB than those with I or II (20% vs. 6%, p-value=0.2956). A complete summary of the bivariate associations is presented in Table 2.

Table 1: Summary of Risk for Sleep-Disordered Breathing, Risk of Emotional/Behavioral Problems, and Intraoral Measures for a Sample of Orthodontic Patients

| | | n | % |
|--|-----------------|----|-----|
| Validated Instruments | | | |
| High Risk for Sleep-Disordered Breathing | | | |
| | Yes | 18 | 18% |
| | No | 82 | 82% |
| High Risk for Emotional/Behavioral Problems | | | |
| | Yes | 10 | 10% |
| | No | 90 | 90% |
| Intraoral Measures | | | |
| Ankyloglossia | | | |
| | Normal (>16 mm) | 87 | 87% |
| | I (12-16 mm) | 11 | 11% |
| | II (8-11 mm) | 2 | 2% |
| Tonsil Grade | | | |
| | 0 | 3 | 3% |
| | 1 | 59 | 59% |
| | 2 | 23 | 23% |
| | 3 | 15 | 15% |
| Friedman Classification | | | |
| | I | 3 | 3% |
| | II | 14 | 14% |
| | III | 50 | 50% |
| | IV | 33 | 33% |
| Palatal Vault | | | |
| | Low | 4 | 4% |
| | Medium | 75 | 75% |
| | High | 21 | 21% |
| Tooth Wear | | | |
| | Mild | 92 | 92% |
| | Moderate | 8 | 8% |
| | Severe | 0 | 0% |
| Spread of Wear | | | |
| | Incisal | 43 | 43% |
| | Posterior | 15 | 15% |
| | Generalized | 29 | 29% |
| | Not Documented | 13 | 13% |
| Molar Classification | | | |
| | I | 61 | 61% |
| | II | 30 | 30% |
| | III | 9 | 9% |
| Skeletal Classification | | | |
| | I | 66 | 66% |
| | II | 23 | 23% |
| | III | 11 | 11% |
| Crossbite | | | |
| | None | 89 | 89% |
| | Unilateral | 9 | 9% |
| | Bilateral | 2 | 2% |

Table 2: Bivariate Associations with High Risk for Sleep-Disordered Breathing

| | | High Risk for SBD | | |
|--|-----------------|-------------------|---------|---------|
| | | Yes | No | P-value |
| Ankyloglossia | | | | 0.4532 |
| | Normal (>16 mm) | 17, 20% | 70, 80% | |
| | I, II (8-16mm) | 1, 8% | 12, 92% | |
| Tonsil Grade | | | | 0.2468 |
| | 0/1 | 9, 15% | 53, 85% | |
| | 2 | 4, 17% | 19, 83% | |
| | 3 | 5, 33% | 10, 67% | |
| Friedman Classification | | | | 0.2956 |
| | I/II | 1, 6% | 16, 94% | |
| | III/IV | 17, 20% | 66, 80% | |
| Palatal Vault | | | | 0.1533 |
| | Low | 2, 50% | 2, 50% | |
| | Medium | 14, 19% | 61, 81% | |
| | High | 2, 10% | 19, 90% | |
| Tooth Wear | | | | 0.6323 |
| | Mild | 16, 17% | 76, 83% | |
| | Moderate | 2, 25% | 6, 75% | |
| Spread of Wear | | | | 0.2429 |
| | Incisal | 7, 16% | 36, 84% | |
| | Posterior | 1, 7% | 14, 93% | |
| | Generalized | 8, 28% | 21, 72% | |
| Molar Classification | | | | 0.7215 |
| | I | 12, 20% | 49, 80% | |
| | II | 4, 13% | 26, 87% | |
| | III | 2, 22% | 7, 78% | |
| Skeletal Classification | | | | 0.6956 |
| | I | 11, 17% | 55, 83% | |
| | II | 4, 17% | 19, 83% | |
| | III | 3, 27% | 8, 73% | |
| Crossbite | | | | 0.3622 |
| | None | 18, 20% | 71, 80% | |
| | Unilateral | 0, 0% | 9, 100% | |
| | Bilateral | 0, 0% | 2, 100% | |
| High Risk for Emotional/Behavioral Problems | | | | <0.0001 |
| | Yes | 8, 80% | 2, 20% | |
| | No | 10, 11% | 80, 89% | |

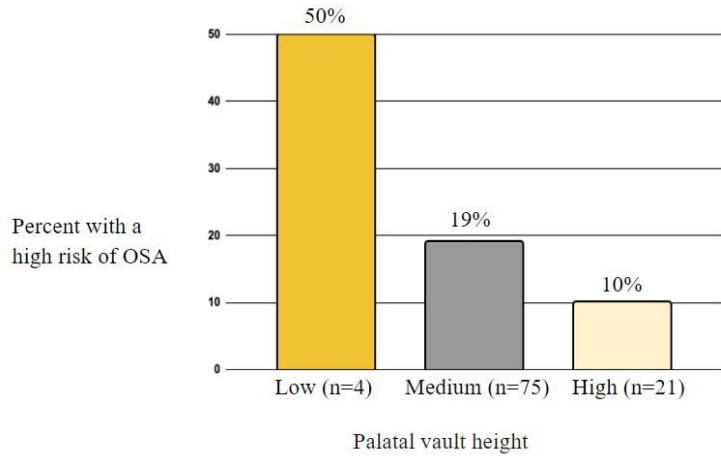


Figure 5: Palatal vault height and risk of OSA

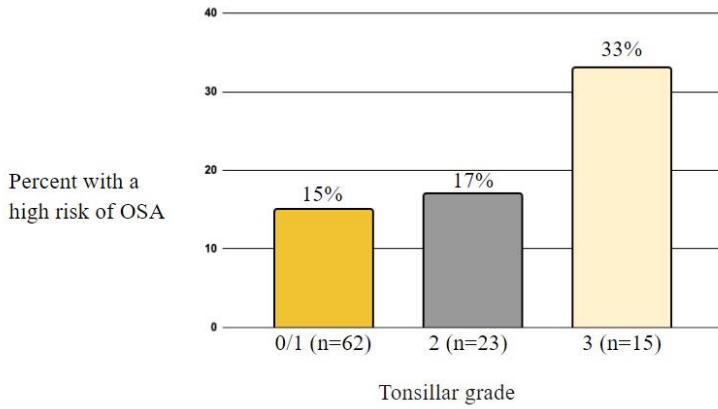


Figure 6: Tonsillar grade and risk of OSA

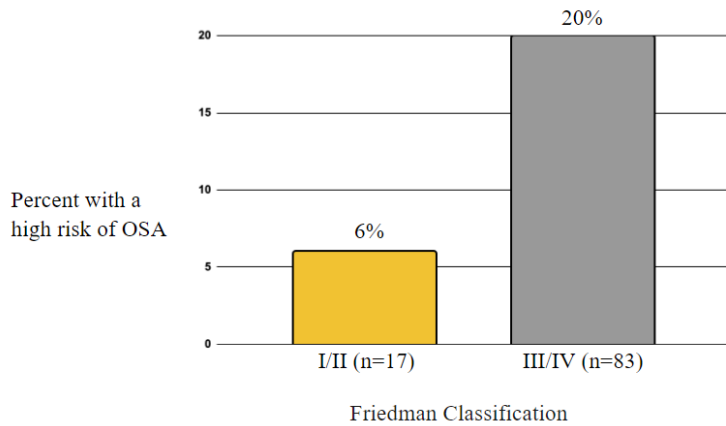


Figure 7: Friedman classification and risk of OSA

Discussion

Over the past several years, sleep apnea has received more attention, particularly in orthodontics. Sleep-disordered breathing in children is a serious health concern that often goes undiagnosed. With a deep understanding of orofacial growth, development, and anatomy in their youth and adolescent patients, orthodontic specialists are uniquely positioned to recognize signs of pediatric OSA and screen for individuals who may benefit from a sleep medicine referral.

While most orthodontists are aware of potential airway issues in their patient population, many still do not routinely evaluate for sleep-disordered breathing in their offices. According to a recent survey by Triggs et al.,²⁵ only 61.5% of practicing orthodontists in the United States screen for OSA. A lack of confidence by the practitioner in their ability to screen for OSA was indicated as the reason for low participation.²⁵ Similarly, 70% of pediatric dentists performing routine screening in their practice report a lack of confidence regarding their capability for OSA evaluation.²⁶ The availability of a simple intraoral examination guideline could be helpful for dental professionals to implement a protocol for the evaluation of OSA in their pediatric patients. Since clinical examination already includes the review of intraoral features, the additional steps for OSA screening can be easily incorporated into routine dental practice. This study evaluated nine intraoral characteristics to determine if they were good predictors for identifying children at risk for OSA.

In the current study, 18% of the screened children were identified as at high risk for sleep-disordered breathing, which was higher than the previously reported prevalence of 7-11%.^{7,8} The variation in the prevalence may be due to differences in patient ages, inclusion criteria, and site selection among the studies. A higher prevalence in the current study could also indicate increased sleep-disordered breathing in the orthodontic patient population. Based on the results of this study and previously reported prevalence data, it is plausible to assume that with millions of children undergoing orthodontic treatment, many orthodontic patients may be suffering from this disorder.

The PSC screening tool for behavioral health problems indicated that ten out of 100 patients were at risk, which agrees with previous reports.²⁷ A high prevalence of behavioral problems may suggest that many orthodontic patients have emotional issues. While depression, anxiety, and other psychological conditions are not within the scope of orthodontics, it is still important to bear in mind that mental health issues are common in young orthodontic patients, which may require referral to specialists.^{28,29}

Anklyoglossia:

Yoon et al.³⁰ reported an average lingual frenulum length between 16-17 mm for 576 children and adolescents ages 6-17 years old, ranging from 6-35 mm, but did not report prevalence data for individuals with frenulum over 16 mm. Villa et al.³¹ noted an average length between 18-19 mm for 504 children ages 6-14 years old in a school in Rome, with 22.6% having a short frenulum. Similarly, Brozek-Mądry et al.³² determined an average of 22 mm for 135 children ages 3-17 years, with 14.8% having a short frenulum.³² In the current study, frenulum length over 16 mm was considered normal. Ratings of I and II corresponding to 8-11 mm and 12-16 mm indicated restriction of the tongue's range of motion. Thirteen subjects had

ankyloglossia in the range of 8-16 mm for the frenulum length (13%). It should be noted that due to differences in the scales used to determine ankyloglossia, it is difficult to compare the studies. Only one of the 18 subjects deemed high-risk for OSA demonstrated ankyloglossia, and the association between the risk of OSA and frenulum length was insignificant ($p=0.4532$).

Tonsillar Grade:

According to Li et al.,²⁰ tonsil size is positively correlated with pediatric OSA, but this study was based on lateral neck radiographs and only had a sample size of 35 children who were referred for suspected OSA. While large tonsils may be the leading cause of pediatric sleep apnea, based on our results, it may not be a strong predictor for screening for pediatric OSA or may simply require a higher power to detect. In this investigation, five out of fifteen (33%) participants with a Tonsillar Grade of 3 were at increased risk for SBD compared to only nine out of 62 (15%) and four out of 23 (17%) for those with a grade of 0/1 or 2, respectively (p -value=0.2468). While there is a positive trend towards a higher risk of OSA with a greater tonsillar grade, the differences were not statistically significant. A more recent study by Kang et al.³³ with 495 symptomatic children found that intraoral determination of tonsil grade was positively related to the apnea-hypopnea index for children ages 1-18, suggesting further investigation for tonsillar grade as a predictor of pediatric OSA.

Friedman's Classification:

Ingram et al.²² failed to find an association between tongue position and SDB in children. In our study, participants with Friedman Classifications of III or IV, indicating higher levels of obstruction at the hypopharyngeal level, demonstrated a higher risk for SBD (17 out of 83, 20%, were deemed high-risk for OSA) compared to those with a Friedman Classification of I or II

(only one out of 17, 6%, were considered high-risk for OSA). The result, however, was not statistically significant ($p=0.2956$).

Palatal Vault:

In the literature, the correlation between palatal height and airway resistance has been previously evaluated.³⁴⁻³⁶ However, the results are conflicting, and there is no reliable evidence that palatal anatomy affects breathing during sleep.³⁷⁻⁴⁰ Also, measurements in previous studies were often carried out on intraoral scans, lateral cephalograms, or casts which do not mimic the routine initial exam. In the current study, the data suggest that a low palatal vault height may be associated with a higher risk for pediatric OSA since two out of four subjects (50%) with a low palatal vault had an increased risk of OSA compared to only 14 out of 75 subjects (19%) with a medium palatal vault and two out of 21 subjects (10%) with a high palatal vault. Decreased space for the tongue may be the cause for reduced upper airway patency. It is important to note that with only four individuals reported to have a low palatal vault height in our sample and a p-value of 0.1533, this should be interpreted with caution.

Tooth wear:

Sleep bruxism is an oral condition associated with obstructive sleep apnea.^{41,42} Bruxism may contribute to tooth wear, which can be easily diagnosed by dentists. A strong association between sleep apnea and tooth wear has been previously reported.⁴¹⁻⁴³ Duran Cantolla et al.⁴³ noted a significant association between apnea-hypopnea index and tooth wear in adult patients. In contrast, the results of this study showed that only 2 out of 18 subjects at high-risk presented with significant wear, with the remaining 18 individuals with mild to no wear. The association between tooth wear and risk of OSA was not statistically significant, with a p-value of 0.6323.

Skeletal/Molar Classification:

Previous literature regarding OSA has investigated airway dysfunction and the Class II hyperdivergent phenotype.¹⁵ In this study, however, when analyzing which malocclusion had the highest prevalence of subjects who scored high-risk for OSA, Class II had the lowest prevalence for both skeletal and dental classifications. The p-values for the association between high risk of OSA and classification indicated no significant relationship (p=0.6956 for skeletal classification and p=0.7215 for molar classification), implying that both skeletal and dental classification do not have strong predictive power when it comes to screening for pediatric OSA.

Crossbite:

A narrow maxilla and a posterior crossbite have been associated with pediatric OSA.¹⁶ In fact, studies have shown that rapid maxillary expansion treatment may even reduce the mean apnea-hypopnea index in children with OSA.^{45,46} However, none of the 18 subjects at high risk for sleep apnea had a crossbite in our sample. All 11 subjects with unilateral or bilateral crossbite had PSQ scores that deemed them at low risk for OSA. Similar to the Class II relationships, while it has been suggested that OSA may lead to the development of a narrow maxilla, the reverse, namely a narrow maxilla indicating OSA, does not appear to hold true (p=0.3622). This is consistent with a recent study by Abulhamayel.⁴⁶

PSC:

SDB in children has been reported to correspond with behavioral issues.^{2,6} When examining the PSQ statistics in relation to the other data collected, there was a strong association between high risk on the PSQ and PSC surveys (p-value<0.0001). This corroborates with previous reports, which stated that preschool children with OSA present significantly impaired executive functions, impaired attention, and more hyperactive behavior problems, emphasizing

the link between OSA and behavioral disturbances.^{47,48} The strong association found in this study was not surprising, and it further supports the importance of investigating pediatric OSA also to alleviate the behavioral issues in young patients afflicted with OSA.⁴⁹

While none of the intraoral markers demonstrated statistically significant associations with sleep-disordered breathing as measured by the PSQ, several of these results still illustrate the possibility of clinical relevance, including palatal vault height, Friedman's classification, and tonsillar grade. Due to the low prevalence of SDB and various intraoral measures with values that deviated from normal, the analysis is underpowered for detecting statistical significance. Lingual frenulum length, tooth wear, molar classification, skeletal classification, and crossbite failed to show either statistical significance or meaningful clinical trends. However, additional research is needed to elucidate the potential use of these presentations in screening for pediatric OSA.

A recognized limitation in this study includes the reliance on parents to report their child's sleeping behavior. Surveys can be subject to reporting errors. Some parents, especially the parents of older children, may not be aware of their child's behaviors. For example, one of the questions asks, "Does your child have a dry mouth on waking up in the morning?" which would be difficult to answer for a subject who drives him or herself to school independently of their caregiver. Sending each patient for a PSG study would be the most reliable way to determine whether the child indeed suffers from OSA, but there are clear limitations to its utilization in terms of cost and accessibility. The aim of our study was to identify associations between intraoral markers and a higher risk of sleep apnea in children without the reliance on surveys, which could then be used in future studies to analyze further with PSG testing.

The most pertinent limitation, however, is the small sample size. A larger sample size of 240 subjects was determined to be adequate to attain statistical significance for any of the intraoral markers. However, due to the time constraint, this study collected data from 100 patients, which was determined as a minimum to carry out the project. The peak prevalence of OSA is reported to be between ages 2-8 years,⁵⁰ and the population that can benefit from orthodontic treatment generally starts at age 7 years, which reduces the chances of having participants test positive on the PSQ survey. Future studies could consider building upon the sample so far in order to reach a greater number of patients with SDB symptoms and deviations from normal with regards to the intraoral markers and consequently detect intraoral markers with high predictive value.

However, it is possible that no set of intraoral markers can be used as a sole screening tool for pediatric OSA. A patient with a high palatal vault, a class II malocclusion, and posterior crossbite would be seen as a typical OSA patient¹⁶, but there are also numerous young patients without these intraoral findings who have an increased body mass index or a neuromuscular disease and related SDB complications. The epidemic of childhood obesity is occurring at progressively younger ages and having obvious health consequences, including pediatric OSA.⁴⁵ Adenotonsillectomy is the treatment of choice for pediatric OSA, but it is not curative in all obese children with hypertrophied adenoids and tonsils, perhaps due to adipose tissue deposited around the pharynx or subcutaneous tissue of the neck leading to compression of the pharynx.⁴⁶

Due to the large variability in the presentations of children with OSA, there are significant challenges in composing a simple screening tool. The pediatric modified STOP-BANG, presented by Chiang et al.,⁵³ although it has not been validated to use for pediatric OSA and SDB screening, may be a more diagnostically accurate screening tool because it includes

extraoral factors implicated in pediatric OSA, such as body mass index, in addition to intraoral factors that have been shown to be related to pediatric OSA, such as tonsillar hypertrophy.⁵³ Still, the pediatric modified STOP-BANG does not include palatal vault height or Friedman's Classification, which both may prove to be valuable predictors of SDB in children if studied in a larger sample, and perhaps even other intraoral variables that were underpowered in the current study.

It is important to include orthodontists and other dental providers who treat children in an effort to reduce the burden of pediatric OSA. Information we can obtain from studies like this will help us to discern which variables are the most and least effective to include in a pediatric OSA screening tool. More research is required to determine if additional modifications to currently available tools, such as the PSQ and PM-STOP BANG, would prove to have higher predictive value and clinical utility in order to make pediatric OSA screening more accessible and efficacious and therefore help more children with SDB get the help that they need.

Conclusion

According to the current study, 18% of subjects are considered at high risk for OSA based on the Pediatric Sleep Questionnaire. While none of the intraoral markers demonstrated statistically significant associations with sleep-disordered breathing, several variables exhibited important clinical trends and warrant further investigation with a larger sample size.

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