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ORIGINAL RESEARCH

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Pediatric craniofacial risk factors for sleep-disordered breathing in dental and orthodontic practice settings: an observational pilot study

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Abstract

Background: Pediatric sleep-disordered breathing (SDB) is highly prevalent and associated with craniofacial risk factors, including malformations of the maxilla, mandible, and other features not routinely assessed clinically. This study evaluated craniofacial risk factors associated with SDB symptoms in children presenting for routine dental care. **Methods**: Children aged 5–12 years were recruited from pediatric dental practices in nine U.S. cities. The primary outcome was SDB as defined by the Pediatric Sleep Questionnaire—Sleep-Related Breathing Disorder (PSQ-SRBD) questionnaire. Regression analyses modeled the association between PSQ-SRBD, the 21 individual craniofacial features, as well as the total number of craniofacial risk factors. A subset of participants underwent a blinded duplicate craniofacial assessment to evaluate inter-rater reliability. **Results**: The sample included 141 children (75% Non-Hispanic-White, 12% Hispanic, 9% Non-Hispanic-Black, 4% other; 50% female), with a mean age (standard deviation) of 8.6 (2.3) years, presenting for routine dental care. PSQ-SRBD scores were $\geq$0.33, signifying increased SDB risk in 28 (20%) children. Increased risk for SDB was associated with the total number of craniofacial risk factors, odds ratio (OR) of 1.16 (95% confidence interval (CI): 1.01–1.35, p = 0.035); as well as with specific individual features including a narrow palate: 3.47 (1.34–9.07, p = 0.010), open bite: 5.61 (1.36–23.63, p = 0.016), tongue thrust: 11.33 (1.98–75.19, p = 0.007), and a heart-shaped tongue: 7.41 (1.48–39.38, p = 0.014) after adjustment for age, sex, race, and ethnicity. There was moderate inter-rater reliability (kappa statistic: 0.76, 95% CI: 0.63–0.89) in the subset of 54 subjects with duplicate craniofacial assessments. **Conclusions**: Multiple craniofacial features assessed during routine dental visits were associated with an increased risk for pediatric SDB. This study supports the utility and feasibility of systematic craniofacial screening by dental providers to identify children at risk for SDB and facilitate timely referral and intervention.

Keywords

Craniofacial; Dentist; Obstructive sleep apnea; OSA; Pediatric

1. Introduction

Sleep-disordered breathing (SDB) encompasses a broad spectrum of clinical presentations, ranging from primary snoring to obstructive sleep apnea (OSA), and is highly prevalent in children [1]. Pediatric OSA involves repeated episodes of prolonged partial upper airway obstruction and intermittent complete obstruction resulting in hypoxia, hypercarbia, increased work of breathing, and sleep fragmentation [1]. Both the gas exchange abnormalities and multiple arousals resulting from these obstructive events contribute to cardiovascular, respiratory, metabolic, and neurobehavioral morbidity in children [2–4].

It is conservatively estimated that OSA prevalence in children is between 1 and 4%, thus impacting some 730,000 to 3 million children under the age of 18 years, emphasizing that OSA should be considered a common disease of childhood [5–8]. Due to the dearth of pediatric sleep specialists and pediatric sleep labs, pediatric OSA remains markedly underdiagnosed despite its high prevalence. OSA prevalence has also increased considerably in specific pediatric groups. These include Black children and those of Asian descent, children with a history of prematurity, asthma, those with a family history of OSA, children with congenital/chromosomal disorders such as Down syndrome and syndromic craniofacial anomalies (*e.g.*, Pierre Robin sequence, achondroplasia), obese children and adoles-

cents [7, 9, 10]. Furthermore, disparities exist in children, with reported differential rates of prevalence, severity, and clinical outcomes among minority groups and those from low socioeconomic status (SES) backgrounds [9, 11, 12]. For example, Black children often present with more severe OSA, including a greater degree of nocturnal desaturation [13], are more likely to have residual disease following adenotonsillectomy, the first-line therapy for OSA [14, 15], and are more likely to experience complications related to this surgical procedure [9, 16].

While adenotonsillar hypertrophy is a significant contributing risk factor for upper airway obstruction in most children, the etiology of SDB in children is, in fact, multifactorial [12]. Previous reports have identified a constellation of craniofacial features in otherwise healthy children without genetic or syndromic comorbidities that may increase the risk of SDB and reflect anatomical features associated with obstructed breathing. For example, upper airway obstruction during sleep may result from malformations of the maxilla, mandible, and other facial and physical structures [17]. This suggests that traditional surgical approaches such as adenotonsillectomy may not be indicated or may not be sufficient to completely resolve symptoms in some children with SDB [15, 18–20]. Thus, identifying these craniofacial risk factors becomes crucial for expanding the options for optimal treatment, including oral appliances. Dentists and orthodontists are already routinely screening children for orofacial features impacting the upper airway, often at least annually. Therefore, they are well-positioned to recognize pediatric SDB early in childhood and to facilitate timely diagnosis and appropriate treatment interventions.

To optimize this opportunity to identify children at risk for OSA in dental and orthodontic clinical practice, we sought to evaluate craniofacial risk factors for sleep-disordered breathing in collaboration with a group of dental/orthodontic providers (SO, EL, GR) specializing in pediatric sleep-disordered breathing. Our primary objective was to identify craniofacial features associated with risk of SDB based on validated measures of sleep-disordered breathing risk and symptom burden. We also aimed to assess the feasibility of screening for these craniofacial risk factors in the context of routine dental care and to evaluate dental practitioners' perspectives on implementing craniofacial screening as part of routine practice. Finally, we examined the prevalence of craniofacial risk features observed during routine pediatric dental visits.

2. Methods

2.1 Study design

Children were recruited between 2021 and 2024 from nine U.S. cities (Scituate, MA; Northwood, NH; Granger, Indianapolis, IN; Columbia, IL; Leawood, KS; Shreveport, LA; Temecula, CA; Santee, CA) to participate in this cross-sectional study. A convenience sample of healthy children, aged 5–12 years, who presented for routine dental visits was included. Children previously diagnosed with OSA, those with congenital syndromes potentially impacting the upper airway (*i.e.*, Down syndrome, Pierre-Robin sequence, Achondroplasia, Treacher-Collins syndrome, Hunter's/Hurler's syndromes), those who were unable or unwilling to undergo a routine dental exam, and those presenting with an acute dental concern were excluded from the study. The study was approved by the Boston Children's Hospital Institutional Review Board (BCH). Informed consent was obtained from participants' guardians, and assent was obtained from participants ($\geq$7 years old) before enrollment. Data were collected from all the dental practices using the BCH secure web platform, the BCH RedCap® program.

The caregiver provided sociodemographic data for participants, including age (years), sex (male/female), and caregiver-reported race (American Indian or Alaska Native; Asian; Black or African American; Native Hawaiian or Other Pacific Islander; White or Caucasian; More than one race; Unknown or not reported) and ethnicity (Hispanic or Latino/Latina). Due to the paucity of individuals who identified as American Indian, Asian or those with more than one race, these groups were re-categorized as "Other".

Clinical examinations were conducted by licensed pediatric dentists or practitioners at each site. All examiners used a standardized data collection form with operational definitions for each craniofacial feature assessed. Prior to data collection, examiners participated in webinar-based training sessions.

2.2 Outcomes

The primary outcome was sleep-disordered breathing (SDB) risk and symptoms, assessed with a caregiver-reported questionnaire, the Pediatric Sleep Questionnaire—Sleep-Related Breathing Disorder (PSQ-SRBD). The PSQ-SRBD scale is a 22-item questionnaire that measures SDB symptoms (snoring frequency, loud snoring, observed apneas, difficulty breathing during sleep, daytime sleepiness, inattentive or hyperactive behavior, and other pediatric OSA features) and has been validated in children with polysomnography-confirmed SDB. It is commonly used to assess SDB risk and symptoms in pediatric patients [21]. The individual items are scored as "yes" = 1, "no" = 0, and "don't know" = missing. The PSQ score is calculated as the ratio of the sum of "yes" answers to the number of answered questions. Higher scores correspond to increased SDB risk and symptoms, and scores $\geq$0.33 indicate clinically relevant high symptom burden and elevated risk for OSA in children.

We also assessed general sleep disturbances in the participants using a validated questionnaire, the parent-proxy Patient Reported Outcomes Measurement Information System (PROMIS) Pediatric Sleep Disturbances-SF8 (Parent-Proxy) scale [22], a measure of caregiver-reported sleep quality, sleep depth, and restorative sleep issues (*i.e.*, caregiver perceived concerns regarding their child's difficulties falling asleep, staying asleep, and overall problematic sleep). PROMIS scores have a mean of 50 and a standard deviation (SD) of 10. A score of 60 is defined as elevated.

2.3 Exposures

Craniofacial risk factors were assessed based on 21 unique craniofacial clinical characteristics shown to be associated with an increased risk of obstructed breathing in children,

including mandible placement, facial and cranial distortion, posture, palatal morphology, teeth crowding, tongue position, and frenulum length (Fig. 1) [23–28]. Each craniofacial feature was evaluated and documented, with the total risk profile representing the combined presence of these features.

Additionally, practitioner perspectives on craniofacial screening were assessed by surveying the participating providers with the following four questions: (1) Please rate your overall satisfaction with screening for craniofacial features related to sleep-disordered breathing in children; (2) Please rate the ease of evaluating these craniofacial features during routine dental care; (3) Please rate the likelihood that you will incorporate craniofacial screening for SDB into your dental practice; and (4) Please rate the likelihood that dental providers treating children in general would incorporate craniofacial screening for SDB into their dental practice. The responses included not satisfied, somewhat satisfied, and very satisfied for questions 1 and 2, and not likely, somewhat likely, and very likely for questions 3 and 4.

2.4 Statistical analysis

Multiple-group or two-group comparisons were conducted using Fisher's exact test. Univariable and multivariable logistic regression models were used to evaluate the association between elevated PSQ-SRBD (primary outcome) and the 21 unique craniofacial features (independent variables) in separate models. In these multivariable models, we adjusted for age, sex, race, and ethnicity. To assess inter-rater reliability, a subpopulation of participants was evaluated by two independent dental providers from the same practice who were blinded to each other's ratings. Cohen's kappa statistic was calculated to assess agreement between examiners for categorical craniofacial features. Kappa values were interpreted using standard guidelines: <0.20 indicating slight agreement, 0.21–0.40 fair agreement, 0.41–0.60 moderate agreement, 0.61–0.80 substantial agreement, and 0.81–1.00 almost perfect agreement. Ninety-five percent confidence intervals were calculated for all kappa statistics.

Practitioner perspectives on craniofacial screening were assessed by surveying the participating providers. All tests were two-sided at a significance level of 0.05. Statistical analysis was conducted using R software (Version 4.2.1).

3. Results

3.1 Study participant characteristics

The analytic sample included 141 children (12% Hispanic, 9% non-Hispanic, Black or African American, 75% non-Hispanic White, and 4% Non-Hispanic Other) with a mean age (SD) of 8.6 (2.3) years. Fifty percent of the participants were female. Characteristics of study participants are presented in Table 1.

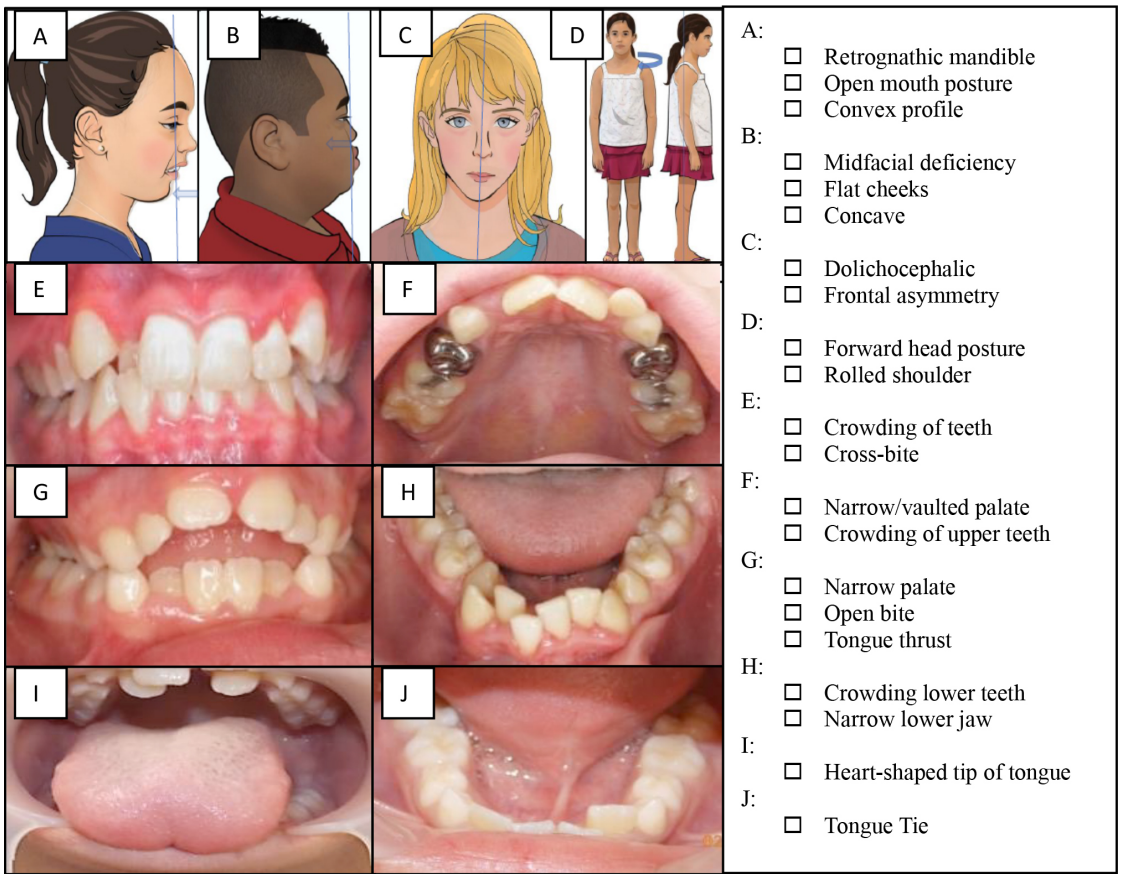

FIGURE 1. Pediatric craniofacial screening tool for sleep (PCSS). The PCSS includes 21 unique craniofacial features associated with an increased risk of obstructed breathing in children and includes mandible placement, facial and cranial distortion, posture, palatal morphology, teeth crowding, tongue position, and frenulum length. The total score is the arithmetic sum of craniofacial features.

TABLE 1. Sample characteristics, overall and by elevated Pediatric Sleep Questionnaire—Sleep-Related Breathing Disorder total score, among children in the study.

	Overall (N = 141)	PSQ-SRBD[a]		*p*-value
		≤0.33 (N = 113)	>0.33 (N = 28)	
Age, mean (SD)	8.6 (2.3)	8.7 (2.3)	8.23 (2.3)	0.281
Sex, n (%)				
Female	71 (50.4)	59 (52.2)	12 (42.9)	0.405
Male	70 (49.6)	54 (47.8)	16 (57.1)	
Race and Ethnicity, n (%)[b]				
White	106 (75.2)	88 (77.9)	18 (64.3)	0.224
Hispanic	17 (12.1)	11 (9.7)	6 (21.4)	
Black	12 (8.5)	10 (8.8)	2 (7.1)	
Other	6 (4.2)	4 (3.6)	2 (7.1)	
PROMIS-Sleep Disturbance T-score[c]				
Mean (SD)	51.7 (9.0)	49.4 (7.9)	61.2 (7.3)	<0.001
≥60, n (%)	23 (16.5)	8 (7.1)	15 (55.6)	<0.001

PSQ-SRBD: Pediatric Sleep Questionnaire-Sleep-Related Breathing Disorder; SD: standard deviation; PROMIS: Patient Reported Outcomes Measurement Information System.
[a]PSQ-SRBD scores range from 0 to 1, with higher scores indicating greater severity of SDB symptoms. A score of 0.33 or greater suggests increased symptoms and risk for a pediatric sleep-related breathing disorder.
[b]Race and ethnicity were based on caregiver responses to categories provided in a questionnaire.
[c]PROMIS-Sleep Disturbance T-scores range from 0–100, with higher scores indicating greater sleep disturbances. A T-score of 60 or greater suggests is 1 SD above the mean and indicative of increased symptoms.
Continuous variables were compared using Wilcoxon rank-sum tests; categorical variables were compared using Fisher's exact tests.

Overall, PSQ-SRBD scores were elevated (≥0.33) in 28 (20%) children and did not differ by covariates age, sex, race, and ethnicity (Table 1). Table 2 describes craniofacial features by race and ethnicity. Craniofacial features, including a dolichocephalic (long face) and narrow, vaulted palate, were more prevalent in Black children. Crowding of lower teeth was more prevalent in Hispanic children. Black children also had a higher number of craniofacial risk features compared with the other groups. The mean number of craniofacial risk features and SD were 7.1 (2.4) in Black, 6.1 (2.3) in Hispanic, 4.6 (3.5) in White, and 5.3 (3.9) in other children.

3.2 Associations between elevated SRBD and craniofacial features

The following individual craniofacial features, as well as the total number of craniofacial risk features, were associated with elevated PSQ-SRBD scores: narrow palate, open bite, tongue thrust, and heart-shaped tongue (Table 3). The multivariable model showed a 3.5-fold increase in the odds for elevated SRBD in children with a narrow palate (95% confidence interval (CI): 1.34–9.07, $p = 0.010$). There were 6- and 11-fold increased odds for elevated SRBD scores in children with open bite (95% CI: 1.36–23.63, $p = 0.016$) and tongue thrust (95% CI: 1.98–75.19, $p = 0.007$), respectively. Additionally, there was 7-fold increase in the odds of elevated SRBD scores children with a heart-shaped tongue (95% CI: 1.48–39.38, $p = 0.014$). The total number of craniofacial risk features was associated with 16% increase in the odds for elevated SRBD scores (OR: 1.16; 95% CI: 1.01–1.35, $p = 0.035$) (Table 4).

3.3 Practitioner interrater reliability and satisfaction with the craniofacial screening

Fifty-four subjects underwent second craniofacial assessments with a kappa statistic of 0.76 (95% CI: 0.63, 0.89) for total score and moderate to substantial kappa values for all significant features, including midfacial deficiency (kappa statistic: 0.60; 95% CI: 0.31, 0.89), crowding of upper teeth (0.62; 0.42, 0.82), crowing of teeth (0.74; 0.56, 0.92), tongue thrust (0.73; 0.38, 1.00), tongue tie (0.76; 0.53, 0.98) and heart-shaped tongue (0.85; 0.55, 1.00) (Table 5).

TABLE 2. Individual craniofacial features, overall and by race and ethnicity, among children in the study.

	Overall (N = 141)	Race and Ethnicity[a]				
		White (N = 106)	Hispanic (N = 17)	Black (N = 12)	Other (N = 6)	*p*-value
	Craniofacial features, n (%)					
Facial						
Retrognathic mandible	48 (34.0)	34 (32.1)	8 (47.1)	5 (41.7)	1 (16.7)	0.488
Open mouth posture	11 (7.8)	8 (7.5)	2 (11.8)	1 (8.3)	0 (0)	0.835
Convex profile	52 (36.9)	43 (40.6)	5 (29.4)	3 (25.0)	1 (16.7)	0.500
Midfacial deficiency	15 (10.6)	10 (9.4)	4 (23.5)	1 (8.3)	0 (0)	0.321
Flat cheeks	41 (29.1)	29 (27.4)	4 (23.5)	7 (58.3)	1 (16.7)	0.141
Concave profile	11 (7.8)	8 (7.5)	2 (11.8)	1 (8.3)	0 (0)	0.835
Dolichocephalic long face	23 (16.3)	13 (12.3)	1 (5.9)	7 (58.3)	2 (33.3)	<0.001
Frontal asymmetry	19 (13.5)	17 (16.0)	2 (11.8)	0 (0)	0 (0)	0.524
Posture						
Forward head posture	63 (44.7)	41 (38.7)	12 (70.6)	7 (58.3)	3 (50.0)	0.062
Rolled shoulders	53 (37.6)	40 (37.7)	6 (35.3)	6 (50.0)	1 (16.7)	0.636
Maxilla/upper jaw						
Narrow, vaulted palate	42 (29.8)	22 (20.8)	8 (47.1)	9 (75.0)	3 (50.0)	<0.001
Crowding upper teeth	40 (28.4)	28 (26.4)	7 (41.2)	3 (25.0)	2 (33.3)	0.618
Mandible/lower jaw						
Crowding lower teeth	63 (44.7)	40 (37.7)	12 (70.6)	8 (66.7)	3 (50.0)	0.026
Narrow lower jaw	33 (23.4)	20 (18.9)	5 (29.4)	5 (41.7)	3 (50.0)	0.084
Maxilla/mandible						
Crowding of teeth	59 (41.8)	41 (38.7)	8 (47.1)	7 (58.3)	3 (50.0)	0.506
Cross-bite	15 (10.6)	11 (10.4)	2 (11.8)	1 (8.3)	1 (16.7)	0.883
Narrow palate	33 (23.4)	21 (19.8)	6 (35.3)	5 (41.7)	1 (16.7)	0.179
Open bite	10 (7.1)	8 (7.5)	0 (0)	1 (8.3)	1 (16.7)	0.421
Tongue thrust	7 (5.0)	6 (5.7)	0 (0)	0 (0)	1 (16.7)	0.372
Tongue						
Tongue tie	41 (29.1)	28 (26.4)	6 (35.3)	4 (33.3)	3 (50.0)	0.507
Heart-shaped of tongue	8 (5.7)	5 (4.7)	1 (5.9)	2 (16.7)	0 (0)	0.267
	Total number of craniofacial risk factors					
Mean (SD)	5.0 (3.4)	4.6 (3.5)	6.1 (2.3)	7.1 (2.4)	5.3 (3.9)	<0.001
Median (IQR)	5 (5)	4 (4)	6 (2)	7 (3)	5.5 (6)	<0.001

SD: standard deviation; IQR: Interquartile Range.
Race and ethnicity were caregiver-reported.
[a] Values presented in the table include the number of overall participants with the described craniofacial features by each racial/ethnic category.
Fisher's exact test was used for multiple-group comparison.

TABLE 3. Individual craniofacial features, overall and by increased risk for sleep-disordered breathing (PSQ-SRBD total score >0.33), among children in the study.

	Overall (N = 141)	PSQ-SRBD		
		≤0.33 (N = 113)	>0.33 (N = 28)	*p*-value
	Craniofacial features, n (%)			
Facial				
Retrognathic mandible	48 (34.0)	38 (33.6)	10 (35.7)	0.827
Open mouth posture	11 (7.8)	7 (6.2)	4 (14.3)	0.229
Convex profile	52 (36.9)	41 (36.3)	11 (39.3)	0.828
Midfacial deficiency	15 (10.6)	11 (9.7)	4 (14.3)	0.498
Flat cheeks	41 (29.1)	36 (31.9)	5 (17.9)	0.169
Concave profile	11 (7.8)	9 (8.0)	2 (7.1)	1.000
Dolichocephalic long face	23 (16.3)	18 (15.9)	5 (17.9)	0.779
Frontal asymmetry	19 (13.5)	13 (11.5)	6 (21.4)	0.214
Posture				
Forward head posture	63 (44.7)	47 (41.6)	16 (57.1)	0.202
Rolled shoulders	53 (37.6)	39 (34.5)	14 (50.0)	0.190
Maxilla/upper jaw				
Narrow, vaulted palate	42 (29.8)	31 (27.4)	11 (39.3)	0.251
Crowding upper teeth	40 (28.4)	29 (25.7)	11 (39.3)	0.166
Mandible/lower jaw				
Crowding lower teeth	63 (44.7)	52 (46.0)	11 (39.3)	0.672
Narrow lower jaw	33 (23.4)	26 (23.0)	7 (25.0)	0.807
Maxilla/mandible				
Crowding of teeth	59 (41.8)	47 (41.6)	12 (42.9)	1.000
Cross-bite	15 (10.6)	11 (9.7)	4 (14.3)	0.498
Narrow palate	33 (23.4)	21 (18.6)	12 (42.9)	0.011
Open bite	10 (7.1)	5 (4.4)	5 (17.9)	0.027
Tongue thrust	7 (5.0)	3 (2.7)	4 (14.3)	0.029
Tongue				
Tongue tie	41 (29.1)	32 (28.3)	9 (32.1)	0.817
Heart-shaped of tongue	8 (5.7)	4 (3.5)	4 (14.3)	0.050
	Total number of craniofacial risk factors			
Mean (SD)	5.0 (3.4)	4.8 (3.2)	6.1 (3.8)	<0.001
Median (IQR)	5 (5.0)	4 (5.0)	6 (3.25)	<0.001

PSQ-SRBD: Pediatric Sleep Questionnaire-Sleep-Related Breathing Disorder; SD: Standard deviation; IQR: Interquartile Range.
Continuous variables were compared using Wilcoxon rank-sum tests; categorical variables were compared using Fisher's exact tests.

TABLE 4. Odds ratios (95% CI) for the associations between craniofacial features and increased risk for sleep-disordered breathing symptoms (PSQ-SRBD total score >0.33).

	Unadjusted model		Adjusted model[a]	
	OR (95% CI)	*p*-value	OR (95% CI)	*p*-value
Craniofacial features				
Facial				
Retrognathic mandible	1.10 (0.45, 2.57)	0.835	1.04 (0.41, 2.53)	0.929
Open mouth posture	2.52 (0.62, 9.07)	0.165	3.19 (0.72, 12.95)	0.106
Convex profile	1.14 (0.47, 2.64)	0.768	1.45 (0.58, 3.59)	0.415
Midfacial deficiency	1.55 (0.40, 4.97)	0.487	1.32 (0.32, 4.55)	0.673
Flat cheeks	0.46 (0.15, 1.24)	0.151	0.44 (0.13, 1.24)	0.143
Concave profile	0.89 (0.13, 3.71)	0.885	0.89 (0.13, 3.96)	0.894
Dolichocephalic long face	1.15 (0.35, 3.23)	0.805	1.29 (0.35, 4.19)	0.681
Frontal asymmetry	2.10 (0.68, 5.96)	0.175	2.25 (0.68, 6.87)	0.164
Posture				
Forward head posture	1.87 (0.82, 4.40)	0.142	1.75 (0.72, 4.31)	0.215
Rolled shoulders	1.90 (0.82, 4.41)	0.133	2.22 (0.92, 5.45)	0.076
Maxilla/upper jaw				
Narrow, vaulted palate	1.71 (0.71, 4.03)	0.223	1.69 (0.62, 4.48)	0.296
Crowding upper teeth	1.87 (0.77, 4.44)	0.156	1.75 (0.70, 4.32)	0.224
Mandible/lower jaw				
Crowding lower teeth	0.76 (0.32, 1.75)	0.522	0.62 (0.24, 1.54)	0.315
Narrow lower jaw	1.12 (0.40, 2.82)	0.824	1.10 (0.37, 2.99)	0.861
Maxilla/mandible				
Crowding of teeth	1.05 (0.45, 2.42)	0.903	0.98 (0.40, 2.37)	0.967
Cross-bite	1.55 (0.40, 4.97)	0.487	1.87 (0.46, 6.50)	0.341
Narrow palate	3.29 (1.34, 7.99)	0.008	3.47 (1.34, 9.07)	0.010
Open bite	4.70 (1.22, 18.20)	0.022	5.61 (1.36, 23.63)	0.016
Tongue thrust	6.11 (1.27, 32.75)	0.023	11.33 (1.98, 75.19)	0.007
Tongue				
Tongue tie	1.20 (0.47, 2.87)	0.690	1.26 (0.47, 3.20)	0.634
Heart-shaped of tongue	4.54 (1.01, 20.47)	0.041	7.41 (1.48, 39.38)	0.014
Total number of craniofacial risk factors	1.13 (1.00, 1.29)	0.050	1.16 (1.01, 1.35)	0.035

OR: odds ratio; CI: confidence interval.
[a] *Model was adjusted for age, gender, race, and ethnicity.*

TABLE 5. Inter-rater reliability between dental practitioners assessing craniofacial features and the total number of craniofacial risk factors (n = 54).

Craniofacial features	Kappa statistic (95% CI)
Facial	
Retrognathic mandible	0.55 (0.33, 0.77)
Open mouth posture	0.31 (−0.16, 0.78)
Convex profile	0.68 (0.47, 0.88)
Midfacial deficiency	0.60 (0.31, 0.89)
Flat cheeks	0.46 (0.18, 0.73)
Concave profile	0.27 (−0.07, 0.60)
Dolichocephalic long face	0.44 (0.07, 0.81)
Frontal asymmetry	0.48 (0.21, 0.76)
Posture	
Forward head posture	0.43 (0.17, 0.69)
Rolled shoulders	0.46 (0.18, 0.75)
Maxilla/upper jaw	
Narrow, vaulted palate	0.50 (0.13, 0.66)
Crowding upper teeth	0.62 (0.42, 0.82)
Mandible/lower jaw	
Crowding lower teeth	0.62 (0.4, 0.83)
Narrow lower jaw	0.46 (0.20, 0.73)
Maxilla/mandible	
Crowding of teeth	0.74 (0.56, 0.92)
Cross-bite	0.43 (0.13, 0.73)
Narrow palate	0.46 (0.22, 0.71)
Open bite	0.48 (−0.13, 1.00)
Tongue thrust	0.73 (0.38, 1.00)
Tongue	
Tongue tie	0.76 (0.53, 0.98)
Heart-shaped tongue	0.85 (0.55, 1.00)
Total number of craniofacial risk factors	0.76 (0.63, 0.89)

CI: confidence interval.
The analysis included 54 participants who had two screenings performed by two independent dental practitioners.

Sixteen participating dental providers completed the satisfaction survey (Fig. 2). Overall, most of the dental providers were satisfied with craniofacial screening for SDB: not satisfied (n = 1, 6%), somewhat satisfied (n = 7, 44%), and very satisfied (n = 8, 50.0%). Dental providers also reported satisfaction with the ease of using the craniofacial screening for SDB during routine dental care, which was highly rated: not satisfied (n = 0, 0%), somewhat satisfied (n = 5, 31%), and very satisfied (n = 11, 69%).

Finally, overall sleep disturbance, assessed by a PROMIS Sleep Disturbance scale score of $\geq$60, was significantly more prevalent in children with elevated PSQ-SRBD (56% *vs.* 7%; $p < 0.001$).

4. Discussion

This study evaluated the relationship between craniofacial features and measures of sleep-disordered breathing symptoms in a healthy cohort of children presenting for routine dental visits across nine U.S. cities. Our findings were striking in that they revealed not only a high percentage of participants who met the criteria for being at risk for SDB (20%), but also a higher-than-expected prevalence of many craniofacial features associated with SDB in this general sample of children. Specifically, features related to the maxilla/mandible and tongue, including narrow palate, open bite, tongue thrust, and heart-shaped tongue, were associated with an increased risk for SDB

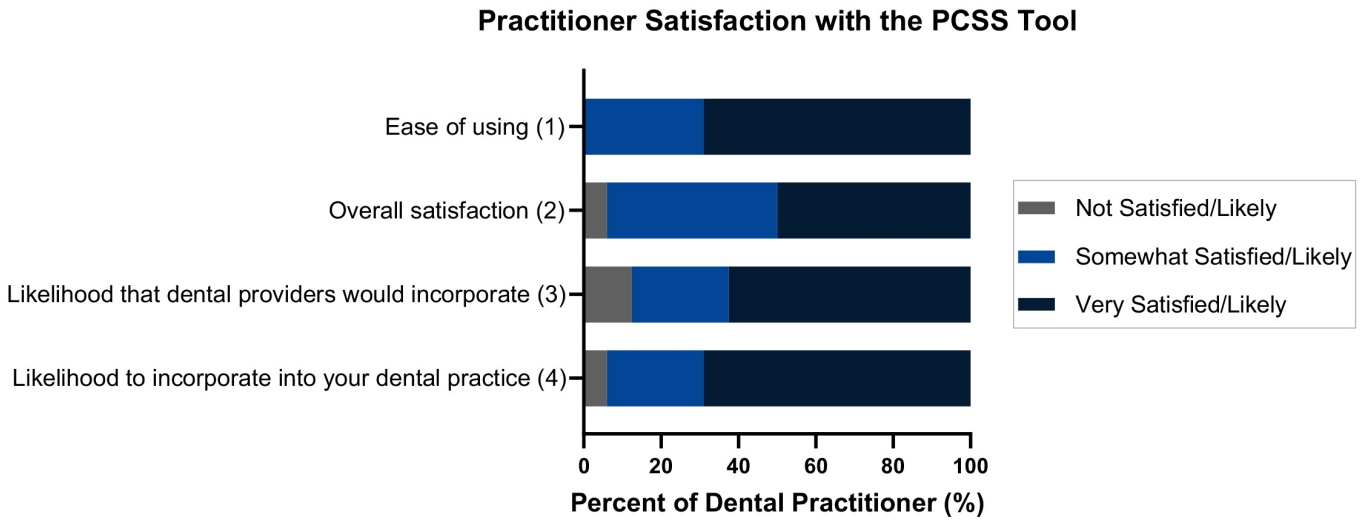

FIGURE 2. Practitioner satisfaction with the PCSS tool. Practitioner satisfaction was assessed using four survey questions. The response selections were not satisfied, somewhat satisfied, and very satisfied for questions 1 and 2, and not likely, somewhat likely, and very likely for questions 3 and 4. PCSS: Pediatric craniofacial screening tool for sleep.

symptoms. There were also racial/ethnic differences in that distinct craniofacial characteristics, including a long, narrow face (dolichocephalic) and narrow vaulted palate, were more prevalent in Black children, while crowding of lower teeth was more prevalent in Hispanic children. Additionally, the craniofacial assessment showed an acceptable level of inter-rater reliability, with 94% of practitioners reporting that they were somewhat or very satisfied with craniofacial screening for SDB, and all practitioners (100%) reporting that they found it somewhat or very easy to perform.

Our findings add to the existing literature by first highlighting the associations between craniofacial features and SDB symptoms in children [26, 29, 30]. The fact that we identified a high prevalence of craniofacial features related to increased SDB risk and symptoms in a group of healthy non-syndromic children is particularly relevant, as previous studies have reported that the vast majority of children with craniofacial malformations have objective evidence of OSA [23–27, 30–32]. Thus, studies that support this association between specific craniofacial features and the risk of SDB justify the routine systematic evaluation and documentation of these craniofacial findings in children within dental practice settings.

Furthermore, since adenotonsillectomy is not indicated for or completely effective as a treatment option for OSA in some children [1, 15, 18, 20, 26], there is an opportunity to encourage the expansion of therapeutic options to include evidence-based non-surgical intervention strategies addressing craniofacial risk factors, such as palatal expansion and mandibular advancement, as well as myofunctional therapy [27]. Thus, the identification, diagnosis, and treatment of sleep-related breathing disorders with oral appliances and other dental/orthodontic treatment modalities are areas where dentists and sleep medicine physicians must work collaboratively to benefit the patients [17]. Moreover, our results also emphasize the need for more epidemiologic data and pediatric randomized controlled trials [31] to identify specific groups of children who may be optimal candidates for combination therapy.

Whereas previous studies have used polysomnography (PSG) to define SDB outcomes [33], our study focused on parent-reported outcomes of SDB symptoms. This was based on the premise that polysomnography may not be widely or easily accessible to dental providers, and that even if available, the PSG apnea/hypopnea index may not fully reflect the clinical manifestations and symptom burden experienced by children with SDB [34]. Moreover, patient/caregiver-reported measures of symptom burden are key outcomes that are increasingly being used in the evaluation of patients [35–37]. Another interesting secondary finding in this study was the high prevalence of subjects with elevated PSQ-SRDB scores who also had increased sleep disturbance scores on the PROMIS scale (56%). This highlights the potential relationship between perceived poor sleep quality as rated by caregivers and SDB symptoms. It also reinforces previous study findings that many children with SDB also have other unrelated sleep disorders, such as insomnia, that require a distinct treatment approach, and thus underscores the need for a systematic and comprehensive evaluation of presenting sleep complaints to identify potential comorbid sleep disorders [38].

In terms of racial/ethnic differences, our study found that Black children more commonly had a high vault palate, which may negatively affect the volume of the nasal cavity and impact normal airflow [31]. Hispanic children were more likely to have tooth crowding, which may reflect a lack of transverse development of the maxilla and mandible, and thus may negatively affect the volume of the airway at the oropharyngeal level, as well as reduce nasal volume [31].

Finally, our study focused on craniofacial features that are potentially associated with increased risk of pediatric SDB because these factors are less frequently recognized contributors to SDB by pediatric providers, including dental practitioners [17, 26]. In addition, dentists and orthodontists are uniquely positioned to identify and refer these children, as they are al-

ready involved in the routine screening of children for orofacial features impacting dentition, alignment, and bite. At the same time, we also acknowledge that it is very important for pediatric clinicians, including dentists, to observe and document the presence of other established SDB risk factors such as obesity and tonsillar hypertrophy as part of a comprehensive approach to screening and diagnosis.

The study and analyses presented here have several strengths. This observational study included a general sample of participants from multiple geographic regions, and used a validated measure to assess sleep symptoms. We used clinical expertise and evidence-based literature to systematically evaluate craniofacial features associated with increased SDB risk. In particular, the prominence in our findings regarding the contribution of tongue position related to a short lingual frenulum ("tongue tie") is noteworthy. Positioning of the tongue on the floor of the mouth is associated with dysfunctional swallowing and mouth breathing [39, 40]. The latter increases the collapsibility of the upper airway and raises the risk of snoring, gasping for air during sleeping, and other symptoms associated with SDB. A "heart-shaped" tongue associated with tongue-tie prevents the elevation of the tongue, causing it to rest away from the palate. However, this intraoral feature remains relatively under-appreciated by clinicians except as a contributing factor to difficulties with breastfeeding in infants and articulation errors in older children, and screening for ankyloglossia as a risk factor for SDB is less frequently employed in the U.S. compared to other countries [30]. This is also relevant as the non-invasive treatment modality of myofunctional therapy (MFT) has some empirical support as an alternative to surgical approaches in ankyloglossia [41]. In general, MFT, especially as an adjunct to other treatments for SDB in both adults and children due to its role in increasing oropharyngeal tone [42], should also be considered by dental providers, especially in the context of a positive screen for these tongue-related craniofacial features [28].

Limitations of this study worth considering include the lack of evaluation for additional SDB risk factors noted above, such as adenotonsillar size, obesity, asthma, allergies, or social determinants of health that may additionally contribute to SDB risk and sleep health disparities. Thus, the omission of these key variables may have impacted the observed associations in our study. As an observational pilot study recruiting children from dental clinics in nine U.S. cities, our convenience sampling strategy introduces potential generalizability concerns, as our sample does not reflect the racial and ethnic diversity of the U.S. pediatric population and limits our ability to examine whether craniofacial-SDB associations differ across populations or to establish race/ethnicity-specific normative values. Misclassification and selection bias are possible, given that our assessment specifically focused on dental and orofacial features clinically observable in the office setting and thus did not include higher-level direct evaluation of deeper craniofacial structures such as cranial base morphology, soft palate dimensions, or three-dimensional upper airway configuration, which are known contributors to SDB pathophysiology and may provide additional explanatory value beyond the features we assessed [43]. The lack of objective sleep testing in this pilot study means our findings reflect associations between craniofacial features and SDB risk or symptoms rather than confirmed SDB diagnoses. Despite this limitation and potential reporting bias, subjective screening tools like the PSQ-SRBD play an important role in clinical practice and research, particularly in large-scale epidemiological studies where PSG is not feasible.

Finally, the study has limitations related to sample size and model complexity. Although multivariable logistic regression models were adjusted for age, sex, race, and ethnicity, the number of children with elevated PSQ-SRBD scores was modest (n = 28), resulting in a low events-per-variable ratio for several analyses. This is particularly relevant for infrequent craniofacial features with small cell counts, which likely contributed to wide confidence intervals and statistical imprecision for some estimates. While craniofacial features were evaluated in separate models to reduce collinearity, residual correlations among related anatomical features cannot be excluded. Accordingly, these findings should be interpreted cautiously and viewed as exploratory, underscoring the need for larger studies with sufficient outcome events to support more stable multivariable modeling.

While the associations we observed between craniofacial features and SDB risk must be interpreted with caution, given the cross-sectional nature of our study, our findings provide valuable preliminary data on craniofacial risk factors that can inform clinical screening practices and guide more targeted diagnostic workups in at-risk populations. Furthermore, the relationship between craniofacial morphology and breathing patterns is likely bidirectional, as certain craniofacial traits (*e.g.*, narrow maxilla, retrognathic mandible) can predispose children to upper airway narrowing leading to obstruction during sleep, chronic mouth breathing, and altered tongue posture, which can subsequently influence craniofacial growth and development. Future studies are needed to evaluate craniofacial screening in larger dental settings with a more racially and ethnically diverse sample of children, while also considering common comorbidities. Additional future outcome measures may include changes in referrals to sleep centers for further evaluation, as well as direct correlation between craniofacial assessment and objective measures of sleep-disordered breathing, including polysomnography.

5. Conclusions

This study demonstrates the utility of systematic craniofacial assessment in children by dental practitioners, particularly about features such as a narrow palate, open bite, tongue position, and lingual frenulum length, to identify children at risk for SDB and selection of appropriate treatment options such as palatal expansion or MFT. Given the profound and consequences of untreated pediatric SDB, adopting a multidisciplinary approach is imperative to improve the likelihood of timely diagnosis and early intervention for affected children. Our findings indicate that craniofacial screening was both reliable and well-accepted by participating dental professionals, supporting its feasibility for implementation in routine dental practice and thereby facilitating appropriate referral and management to improve health outcomes for children with

sleep-disordered breathing.

AVAILABILITY OF DATA AND MATERIALS

The datasets generated and analyzed during the current study are not publicly available due to privacy and confidentiality concerns, but are available from the corresponding author upon reasonable request.

AUTHOR CONTRIBUTIONS

JAO, RB, SRO, EL, and GRY—designed the research study. GKH, ASM, and SGN—performed the research. SGN and BZ—analyzed the data. SGN and JAO—drafted the manuscript. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The study was approved by the Boston Children's Hospital institutional review board (No. IRB-P000038232). Informed consent was obtained from parents or legal guardians of all participating children prior to enrollment.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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