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Authors

Fernandes Fagundes, Nathalia Carolina
Carlyle, Terry
Dalci, Oyku
et al.

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SCIENTIFIC INVESTIGATIONS

Use of facial stereophotogrammetry as a screening tool for pediatric obstructive sleep apnea by dental specialists

Nathalia Carolina Fernandes Fagundes, DDS, MSc[1]; Terry Carlyle, DDS, MSc[1]; Oyku Dalci[2]; M. Ali Darendeliler, DDS, PhD[2]; Ida Kornerup, DDS, MSc[1]; Paul W. Major, DDS, MSc[1]; Andrée Montpetit, DDS, MSc[3]; Benjamin T. Pliska, DDS, MSc[4]; Stacey Quo, DDS, MSc[5]; Giseon Heo, PhD[1,*]; Carlos Flores Mir, DDS, DSc[1,*]

*[1]School of Dentistry, Faculty of Medicine and Dentistry, University of Alberta, Edmonton, Alberta, Canada; [2]Department of Orthodontics, Faculty of Dentistry, University of Sydney, Sydney Dental Hospital, Sydney South West Area Health Service, Sydney, New South Wales, Australia; [3]Department of Oral Health–Orthodontics Section, Faculty of Dental Medicine, Université de Montréal, Montreal, Quebec, Canada; [4]Department of Oral Health Sciences, Faculty of Dentistry, University of British Columbia, Vancouver, British Columbia, Canada; [5]Division of Orthodontics, School of Dentistry, University of California San Francisco, San Francisco, California; *Joint senior authors*

Study Objectives: To evaluate facial 3-dimensional (3D) stereophotogrammetry's effectiveness as a screening tool for pediatric obstructive sleep apnea (OSA) when used by dental specialists.

Methods: One hundred forty-four participants aged 2–17 years, including children fully diagnosed with pediatric OSA through nocturnal polysomnography or at high-risk or low-risk of pediatric OSA, participated in this study. 3D stereophotogrammetry, Craniofacial Index, and Pediatric Sleep Questionnaire were obtained from all participants. Ten dental specialists with interest in pediatric sleep breathing disorders classified OSA severity twice, once based only on 3D stereophotogrammetry and then based on 3D stereophotogrammetry, Craniofacial Index, and Pediatric Sleep Questionnaire. Intrarater and interrater reliability and diagnostic accuracy of pediatric OSA classification were calculated. A cluster analysis was performed to identify potential homogeneous pediatric OSA groups based on their craniofacial features classified through the Craniofacial Index .

Results: Intrarater and interrater agreement suggested a poor reproducibility when only 3D facial stereophotogrammetry was used and when all tools were assessed simultaneously. Sensitivity and specificity varied among clinicians, indicating a low screening ability for both 3D facial stereophotogrammetry, ranging from 0.36–0.90 and 0.10–0.70 and all tools ranging from 0.53–1.0 and 0.01–0.49, respectively. A high arched palate and reversed or increased overjet contributed to explaining how participating dental clinicians classified pediatric OSA.

Conclusions: 3D stereophotogrammetry-based facial analysis does not seem predictive for pediatric OSA screening, alone or combined with the Pediatric Sleep Questionnaire and Craniofacial Index when used by dental specialists interested in sleep-disordered breathing. Some craniofacial traits, more specifically significant sagittal overjet discrepancies and an arched palate, seem to influence participating dental specialists' classification.

Keywords: sleep apnea, obstructive, child, screening, cluster analysis

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BRIEF SUMMARY

Current Knowledge/Study Rationale: One of the main challenges in managing pediatric obstructive sleep apnea is providing a timely diagnosis among this age group. The evaluation of soft tissue facial features among children with obstructive sleep apnea may help identify if specific facial features linked to this morbidity could be used to improve screening algorithms.

Study Impact: This study shows that 3-dimensional stereophotogrammetry-based facial analysis alone or when combined with the Pediatric Sleep Questionnaire and Craniofacial Index does not seem predictive for pediatric obstructive sleep apnea screening. A major sagittal overjet and an arched palate may influence the dental clinician's obstructive sleep apnea classification. The diagnosis and screening for pediatric obstructive sleep apnea should not be oversimplified through excessive reliance on dental specialists' craniofacial features traits.

INTRODUCTION

Obstructive sleep apnea (OSA) is a respiratory sleep disorder resulting in partial or complete airway obstruction.[1] Among children, OSA prevalence has been reported to vary from 1% to 5%.[2,3] In the absence of proper management of OSA cases, commonly a result of underdiagnoses, several health conditions can arise. These include cognitive problems[4] and respiratory and cardiac comorbidities.[5] OSA is related to an increased cost of health services and poor academic progress from a social perspective.[6,7]

Different factors can contribute to pediatric OSA development, such as craniofacial features,[8] tonsils and/or adenoid tissue hypertrophy,[9] reduced upper airway space,[10,11] and obesity.[12] Enlarged tonsils and adenoids are the leading cause of OSA in children.[13] Craniofacial features, more specifically, a high arched palate, convex facial profile, and an anterior open bite, have been linked to OSA in some children.[8]

A key component of pediatric OSA diagnosis is nocturnal polysomnography (nPSG), an exam that monitors oxygen saturation, oronasal airflow, respiratory movement, electroencephalogram, body position, electromyogram, electrooculogram, and

electrocardiogram of a full night of sleep.[14] This exam faces some barriers in many countries, including the high cost and long wait lines for public health services.[15] Alternatively, several screening tools have been used to evaluate pediatric OSA's sleep signs and symptoms,[16] consider adenoid and tonsil sizes,[17] and monitor sleep parameters at home.[18] However, relatively little attention is given to the potential screening of craniofacial features linked to pediatric OSA.

A facial soft tissue analysis as part of a pediatric OSA screening may represent a safe and accessible method for dental professionals' routine clinical use. Dentists and dental specialists are trained to perform facial analysis that are typically incorporated into dental patients' clinical exams. Using craniofacial anthropometry and photogrammetry to evaluate facial features has been proposed as an alternative technique to suggest OSA in adults.[19,20] There is a need to determine if this method would help pediatric OSA screening and how craniofacial features may influence this diagnosis.

Considering some degree of contribution of craniofacial features to the upper airway collapse during sleep, identifying homogenous categories of craniofacial patterns potentially linked to this collapse may help. One of the approaches used to identify these patterns is clinical phenotyping based on clustering methods.[21,22] There is a scarcity of studies exploring the identification of specific craniofacial patterns in pediatric OSA. Among adults, this method has been used to characterize clinical phenotypes in patients with OSA. Adults presenting skeletal Class II hyperdivergent pattern, posteriorly displaced hyoid, and retroclined soft palate were features identified to group moderate to severe patients with OSA.[21]

Understanding the craniofacial feature's role and its evaluation by dental professionals may help identify patients at higher risk for pediatric OSA. This could improve early diagnosis and proper management of OSA in children. This study aims to evaluate the effectiveness of an analysis of facial soft tissue features through stereophotogrammetry as a screening tool for pediatric obstructive sleep apnea by dental specialists.

METHODS

This study was approved by the Health Research Ethics Board of the University of Alberta (Pro00057638). Children and adolescents aged 2–17 years fully diagnosed with pediatric OSA through nPSG or at high-risk or at low-risk for pediatric OSA (normative patients), participated in this study based on Pediatric Sleep Questionnaire (PSQ) score study. The presence of craniofacial syndromes was considered an exclusion criterion.

Children and adolescents under the care of 2 different facilities—a children's hospital sleep center (Pediatric Sleep Laboratory, Stollery Children's Hospital, Edmonton, AB, Canada) and a university's dental clinic (Dental Clinic at the University of Alberta, Edmonton, AB, Canada)—were invited to participate in this study. The participants from the hospital site presented sleep-disordered breathing clinical signs and symptoms and had an nPSG exam. The dental clinic participants were at high-risk or at low-risk for pediatric OSA, assessed by a PSQ questionnaire.

The sample size was calculated based on a type I error rate of 5%, the statistical power of 80%, a null hypothesized value of 0.6, and an alternative hypothesized value of 0.7 for sensitivity and specificity.[16,23] We also set the prevalence rate at 5%. Therefore, the OSA group's minimum required sample sizes are 181 (sensitivity) and 10 (specificity). At the same time, the total sample sizes (both OSA and control group) are 3,620 (sensitivity) and 191 (specificity). However, achieving a total sample size of 3,620 is not realistic, so we set our goal in terms of minimum sample size as an average of sensitivity and specificity for the OSA group, which is approximately 96 and implies that we need to set a total of both OSA group and control group as 192. Consequently, we set the sample size 100 per group and a total of 200.

Nine orthodontists and one pediatric dentist, with a special interest in pediatric sleep disorders, were invited to suggest the potential for pediatric OSA severity on a 4-point ordinal scale (not likely or mild, moderate, severe OSA) among these children based on 3D stereophotogrammetry, Craniofacial Index (CFI)[24] and PSQ.[25] The CFI is a tool developed to identify the orthodontic treatment need in pediatric patients with OSA. This index evaluates the frequency of the 8 most frequent orthodontic problems observed in children with OSA.[24]

The dental specialists have clinical experience in providing dental care to children with sleep-breathing disorders and research interests in pediatric sleep disorders. However, their actual OSA-related knowledge was not directly assessed. This group of clinicians was from Canada, the United States, and Australia.

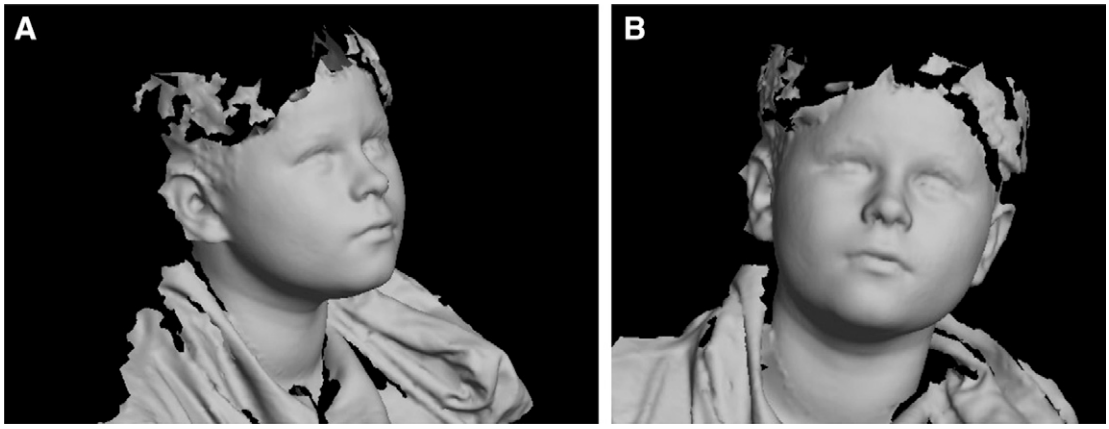
The pediatric sleep questionnaires and craniofacial parameters were collected from all children included in this study. The sex, age, and body mass index (BMI) were collected when available from a subsample of the included children. The BMI z-scores were calculated following the growth standards of the Centers for Disease Control and Prevention. A BMI z-score between 1 and 1.9 indicates overweight, and BMI z-score ≥ 2 indicates obesity.[26]

After nPSG, the Obstructive–Mixed Apnea–Hypopnoea Index (OMAHI) was calculated. This index was calculated based on the number of apneas and hypopneas during sleep divided by the total sleep time, excluding the central respiratory events. Children presenting an OMAHI index ≥ 2 events/h were classified as presenting pediatric OSA.[27] The OSA severity was categorized as mild (OMAHI = 2–4.9 events/h), moderate (OMAHI = 5–9.9 events/h), and severe (OMAHI ≥ 10 events/h).[27]

The PSQ was collected in all 200 participants and nPSG among only 103 participants. 3D stereophotogrammetry was collected in 152 children. Children presenting a PSQ score of ≥ 8 were considered at high-risk for OSA, whereas a PSQ score of < 8 indicated at low-risk for OSA.[25] The 3D facial stereophotogrammetry (3dMD, Atlanta, GA) and the CFI were adopted to evaluate the craniofacial parameters. Facial stereophotogrammetry comprises the estimation of 3D coordinates of facial features utilization of images taken by multiple cameras simultaneously. The cameras are set in different positions around the face. In addition to rendering a 3D image, this data can be utilized to perform anthropometric analyses of facial soft tissue landmarks[28] (**Figure 1A** and **Figure 1B**).

The involved clinicians categorized all children according to each participant's perceived OSA severity as not likely, mild, moderate, or severe in 2 ways. First it was based only on the 3D

Figure 1—Three-dimensional stereophotogrammetry used in the clinician's evaluation.



Lateral **(A)** and frontal **(B)** views.

facial stereophotogrammetry records. After that, it was based on the stereophotogrammetry and additional information obtained from the CFI and the PSQ. Only the total score of PSQ was provided to the clinicians. Only the CFI scores from the intraoral evaluation were available to the evaluators. The clinicians had virtual access to the 3D facial stereophotogrammetry file and rotated and zoomed the images. All clinicians received the same level of instruction regarding the assessment of 3D stereophotogrammetry images. There was no access to the initial severity ranking as determined using the stereophotogrammetry records for this second assessment round.

The intrarater and interrater reliability were calculated among clinicians. The intrarater reliability was evaluated in a subsample of 5 clinicians from the University of Alberta, 4 orthodontists and 1 pediatric dentist, in which Delta was calculated. Only this group was able to evaluate the data twice. The interrater reliability was checked among all 10 clinicians, in which both Delta and Fleiss' Kappa were calculated. Delta was chosen as an alternative to Cohen's Kappa due to the presence of unbalanced marginal totals.[29] The Delta measurement considers the total proportion of answers in agreement and is valid in all circumstances in which Cohen's Kappa is valid.[29] We reported Fleiss' Kappa because it measures the agreement among multiple raters.

The agreement level was considered excellent if above 0.9, good if between 0.75 and 0.9, moderate if in the range of 0.5–0.75, and poor if below 0.5.[30] This agreement level was used for both Delta and Fleiss' Kappa analysis.

The diagnostic value of the classification was evaluated through sensitivity, specificity, positive predictive value (PPV) and negative predictive value (NPV). In addition, the prevalence of pediatric OSA was considered 5% to the PPV and NPV calculation.[2,3] The classification suggested by the participating clinicians was compared to pediatric OSA diagnosis in the group of patients submitted to nPSG.

A cluster analysis was performed to identify and characterize the children's craniofacial features included in this study. This analysis also aimed to understand the relationship between sleep status variables and pediatric OSA classification on children's specific craniofacial features in this sample. A 2-step cluster analysis was performed. First, the 8 craniofacial feature scores evaluated through the Craniofacial Index were entered as unique variables to identify clusters. These scores are representative of the most common craniofacial abnormalities observed in OSA children.[24] The best cluster solution was chosen based on the Akaike Information Criterion and the log-likelihood distance. The number of groups was defined according to the large ratio of Akaike Information Criterion changes and the large ratio of distance measures.

After clustering analysis, a post hoc analysis was performed as follows. The distribution of demographic (sex, age, and BMI z-score) and sleep status variables (PSQ score, diagnosis of pediatric OSA, when available) between clusters were evaluated by descriptive analysis (frequency or mean and standard deviation). We compared the distribution of pediatric OSA classification by all clinicians with clusters. To compare the distribution of OSA classification performed by clinicians across clusters, we combined the classes determined by all 10 clinicians into 1 category by choosing the most frequent classification by all 10 clinicians for each patient. For example, if 10 clinicians' classification for a patient was 1, 1, 2, 2, 3, 4, 2, 2, 3, 4, then most frequent is 2. If the most frequent classes were tied, the lower class was chosen.

We combined 4-category pediatric OSA classification into 2, not likely (previously categorized as not likely) or likely (previously classified as mild, moderate, or severe) for statistical analysis. The SPSS statistical package for the social sciences (version 23; IBM, Armonk, NY) was used for data analysis. A P value < .05 was set as statistical significance.

RESULTS

Among the 200 patients enrolled in the study, 103 participants were recruited from a sleep laboratory and 97 participants from the dental clinic. The 3D stereophotogrammetry was collected in 152 children registered in a sleep laboratory at a pediatric hospital

(nPSG sample, n = 78) and at a university dental clinic (non-nPSG sample, n = 74). The subsample without an nPSG (n = 74) was not considered in the diagnostic evaluation of the 3D stereophotogrammetry screening tool. Even though a complete list of 152 patients was sent to all 10 dental specialists, some missing data were detected during data analysis. In the first categorization performed by all 10 clinicians (only 3D stereophotogrammetry available), 1 patient's evaluation was missing (n = 151). In the second categorization (3D stereophotogrammetry, CFI and PSQ available), 8 evaluations were missing (n = 144). A detailed diagram of participants' flow in the study is presented in **Figure 2**.

In the nPSG sample of participants submitted to the 3D photogrammetry (n = 78), 53% (n = 41) had a positive diagnosis of OSA, including mild to severe cases (**Table 1**). The OSA risk defined by PSQ was high in 76% (n = 59) of the participants, with a mean age of 8.5 ± 4.1 years and a BMI z-score = 0.6 ± 1.6 (n = 65). No strong correlation ($r = .07$, $P = .57$) was observed between PSQ and OMAHI scores (see **Table S1** in the supplemental material). In the group of participants without an nPSG, 13% (n = 20) were at high risk for OSA, with a mean age of 8.9 ± 2.5 years (**Table 1**).

The consistency between 2 trials among 5 University of Alberta clinicians was poor when only 3D stereophotogrammetry was available (Δ= 0.39–0.45) and improved from poor to good (Δ= 0.44–0.75) when 3D stereophotogrammetry, CFI, and PSQ were available (**Table S1**).

Fleiss' Kappa evaluated the agreement among all ten clinicians (**Table S1**). The Fleiss' Kappa was poor in both situations. It was 0.12 when only 3D photos were considered for classification and slightly improved to 0.37 when 3D photos, CFI, and PSQ were assessed. Each clinician's agreement contributed to understanding each clinician's role in the reliability. According to Delta, the agreement was poor to moderate when only the 3D stereophotogrammetry was available (Δ = 0.24–0.53). These results improved when the 3D stereophotogrammetry, PSQ, and CFI were available before pediatric OSA classification in 9 of 10 clinicians, which showed a poor to good reliability (Δ = 0.14–0.86) (**Table S1**). Compared to the other 9 dental specialists, clinician 2 showed a weak performance when all tools were considered together compared to only 3D stereophotogrammetry.

In the first classification of OSA performed by clinicians, only 3D stereophotogrammetry was available. In this scenario, the sensitivity (0.36–0.90) and specificity (0.10–0.56) values presented a large variability among 10 participating clinicians. PPV values varied from 0.05 to 0.07, and NPV varied from 0.95 to 0.98 among 10 clinicians. Among the clinicians, the average values for these measurements were sensitivity = 0.51, specificity = 0.35, PPV = 0.04, NPV = 0.93 (**Table 2**, **Table S2** in the supplemental material).

The second classification of OSA performed by clinicians was based on all tools (3D stereophotogrammetry, PSQ, and CFI). The sensitivity values (0.55–1.0) and specificity (0.01–0.49) increased for 9 of the 10 clinicians than the first classification, but a large variability among the clinicians was still present. The PPV (0.04–0.06) remained very low and NPV (0.87–0.96) very high across the clinicians. Among the clinicians, the average values for these measurements were sensitivity = 0.78, specificity = 0.13, PPV = 0.04, NPV = 0.92 (**Table 2**, **Table S2**).

The 2-step clustering analysis identified 2 different clusters based on the frequency of 8 craniofacial features observed in the sample, identified by the CFI assessment. The clusters presented an acceptable quality (Silhouette's index = 0.5). The cluster's size ratio was 1.5 (cluster A, n = 120 and cluster B, n = 80 children). The most important variables to distinguish clusters were overjet, soft tissue lateral profile, and palate depth. Cluster B presented more children with craniofacial disharmonies linked to pediatric OSA than cluster A. More specifically, cluster B children presented a high arched palate and a significantly increased or reversed overjet, while children in cluster A presented a more normal craniofacial pattern (**Table 3**).

Approximately the same percentage of patients diagnosed with pediatric OSA by nPSG was observed in cluster A (47%) and cluster B (50%). The frequency of children categorized as high risk for OSA, defined after PSQ screening, was slightly higher in cluster B (60%) than cluster A (42%). Cluster B presented more patients categorized as likely to have OSA than cluster A in both clinicians' evaluations. This frequency was higher when the CFI and PSQ were added to the assessment, in which only 37% were classified as likely in cluster A, and 76% of patients were classified as likely in cluster B (**Table 4**).

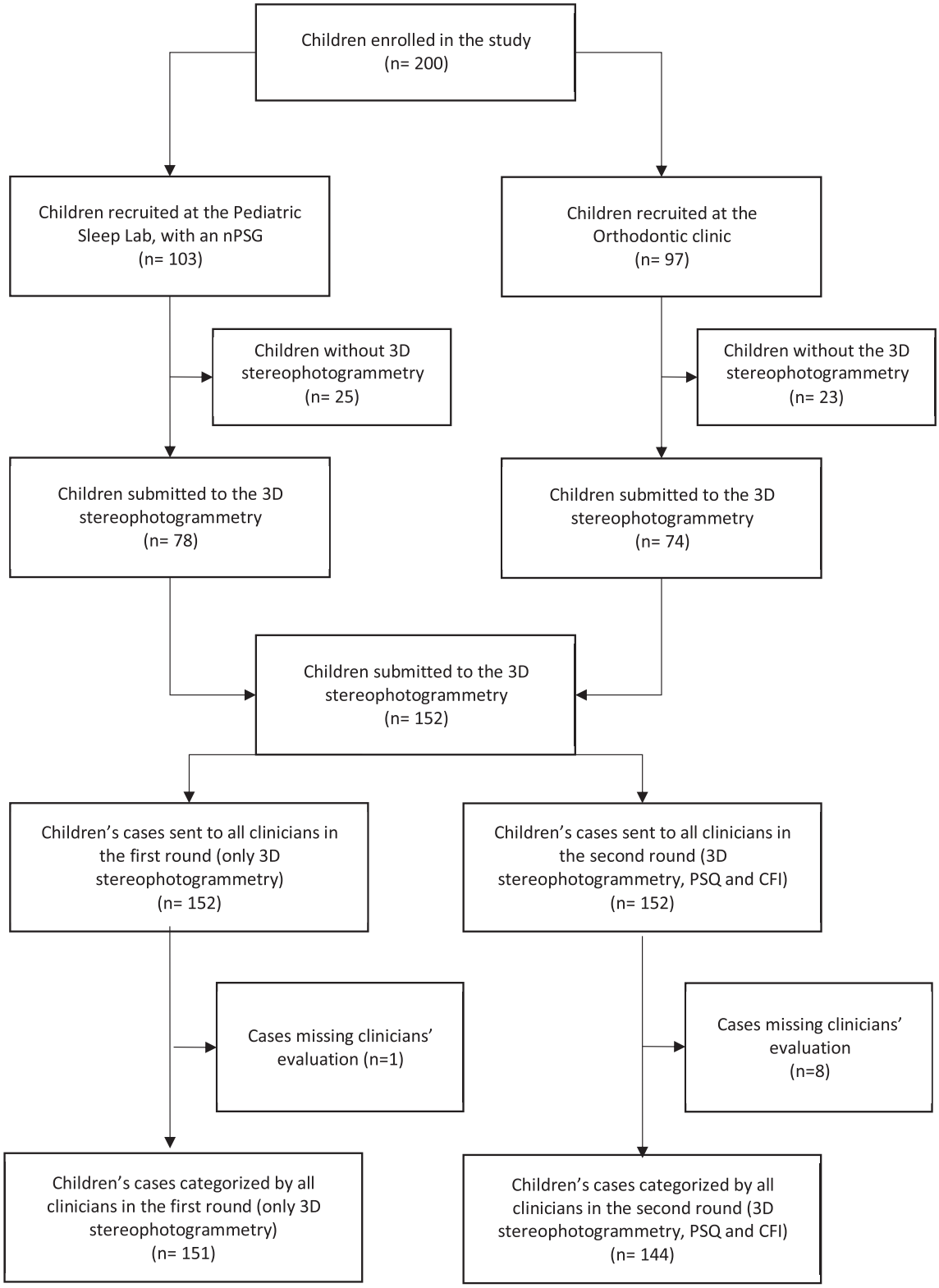
Regarding the distribution of craniofacial features among nPSG children, OSA-negative and OSA-positive patients presented the same magnitude of craniofacial abnormalities (**Table S4** in the supplemental material).

Regarding demographic aspects, only part of the sample reported sex (n = 180), age (n = 196), and BMI z-score (n = 65) due to miscommunication between recruitment sites. In cluster A and cluster B, a balanced ratio of male and female patients was observed. The mean age was higher in cluster B (9.8 ± 3.6 years) than the mean age in cluster A (8.2 ± 3.5 years). The BMI z-score was higher in cluster B (1.1 ± 1.1) than cluster A (0.1 ± 1.9). However, firm conclusions cannot be drawn due to the lack of information for the entire sample.

DISCUSSION

The evaluation of soft facial features by dental specialists based on 3D stereophotogrammetry analysis showed poor intrarater and interrater reliability and low values of sensitivity, specificity, PPV, and a high NPV value among all dental specialists. The availability of additional information about craniofacial features and PSQ scores in addition to the images improved both intrarater and interrater reliability among clinicians but remained questionable for screening purposes. Also, to 9 of 10 clinicians, the sensitivity increased when all tools were assessed, but a negligible specificity was still observed. The presence of significantly reversed or increased overjet, along with a high arched palate, seems to affect how these dental specialists classified patients regarding perceived OSA risk. However, these features appear not to be associated with the final pediatric OSA status evaluated through an nPSG. What this seems to imply is that dental specialists are likely biased by their perception that specific clinical malocclusion traits are highly likely associated with OSA when, in reality, their presence is not expected to imply pediatric OSA automatically. This is an important finding as dental

Figure 2—Flow chart for participants and missing data in the study.



CFI = Craniofacial Index, nPSG = nocturnal polysomnography, PSQ = Pediatric Sleep Questionnaire, 3D = 3-dimensional.

Table 1—Characteristics of participants submitted to 3D stereophotogrammetry.

	nPSG Sample (n = 78)	Non-nPSG Sample (n = 74)
OSA diagnosis		
Negative	37 (47)	NA
Mild	17 (22)	NA
Moderate	20 (26)	NA
Severe	4 (5)	NA
OSA risk evaluated by PSQ		
At low risk	19 (24)	64 (87)
At high risk	59 (76)	10 (13)
Age	8.5 ± 4.1	8.9 ± 2.5
Sex		
Male	41 (52)	39 (53)
Female	37 (48)	35 (47)
BMI z-score	0.6 ± 1.6 (n = 65)	*

Values are presented as frequency, n (%), or mean ± standard deviation. *BMI z-score not available for this subgroup. BMI = body mass index, NA = not applicable, nPSG = nocturnal polysomnography, OSA = obstructive sleep apnea, PSQ = Pediatric Sleep Questionnaire, 3D = 3-dimensional.

Table 2—Sensitivity, specificity, PPV, and NPV of pediatric OSA classification.

Diagnostic Values	Only 3D Stereophotogrammetry: Total (n = 78)	3D Stereophotogrammetry, CFI, and PSQ: Total (n = 75)
Sensitivity (95% CI)	0.51 (0.35, 0.67)	0.78 (0.62, 0.89)
Specificity (95% CI)	0.35 (0.20, 0.52)	0.13 (0.01, 0.28)
PPV (95% CI)	0.04 (0.03, 0.06)	0.04 (0.03, 0.05)
NPV (95% CI)	0.93 (0.89, 0.96)	0.92 (0.80, 0.98)

The average values of sensitivity, specificity, PPV and NPV for all 10 clinicians are shown. CFI = Craniofacial Index, CI = confidence interval, NPV = negative predictive value, OSA, obstructive sleep apnea, PPV = positive predictive value, PSQ = Pediatric Sleep Questionnaire, 3D = 3 dimensional.

clinicians may target a specific subgroup of pediatric OSA patients while potentially ignoring those with OSA but without evident known malocclusion traits.

The evaluation of craniofacial features among children with OSA has been previously explored through cephalometric[8,31] and photographic methods.[32] To our knowledge, this is the first study to evaluate the diagnostic value of 3D facial stereophotogrammetric analysis for pediatric OSA screening. Among adults with OSA, the diagnostic value of craniofacial evaluation by 2-dimensional photographs has been explored by assessing anesthesiologists, otolaryngologists, and internists. Their photo diagnosis has observed a 61.8% accuracy in comparison to nPSG.[33]

In the present study, the facial evaluation using 3D facial stereophotogrammetry by dental specialists with a known interest in sleep-disordered breathing showed considerable variability in the sensitivity and specificity among all 10 clinicians, regardless of the access to PSQ score and CFI index information. The PPV was very low (0.05–0.07), and NPV was high (0.87–0.98) in both classifications.

Overall, when assessing these diagnostic values, it can be concluded that 3D facial stereophotogrammetry without or with the addition of specific craniofacial morphological data was not a valid screening tool for pediatric OSA among this selected group of dental specialists. A moderate number of false positives and a high number of false negatives is suggested in either approach. Significant negative implications could ensue. The false positives will further burden the health system unnecessarily, while the false negatives will deny children the option of being assessed for potential pediatric OSA.

The performance of these screening approaches were lower in comparison to other alternative tools adopted for pediatric OSA screening, such as the PSQ (sensitivity = 0.71 to 0.84 and specificity = 0.13 to 0.72, among 5 studies),[34] overnight oximetry (sensitivity-specificity = 0.80–0.65, 0.85–0.79, and 0.82–0.90 for models classifying children with an apnea-hypopnea index ≥1 events/h, ≥5 events/h, and ≥ 10 events/h, respectively)[35] and Mallampati score (sensitivity = 0.88, 95% confidence interval: 0.80, 0.96; specificity: 0.77, 95% confidence interval: 0.77, 0.68).[17] PSQ is especially useful due to its simplicity

Table 3—Variables used to determine clusters: distribution of craniofacial features across groups.

Craniofacial Features	Cluster A (n = 120)	Cluster B (n = 80)	Total (n = 200)
Overjet			
Normal	120 (100)	24 (30)	144 (72)
Increased or reverse	0 (0)	56 (70)	56 (28)
Profile			
Normal	120 (100)	46 (58)	166 (83)
Severely convex or concave	0 (0)	34 (42)	34 (17)
Palate			
Normal	89 (74)	31 (39)	120 (60)
Mildly high arched	30 (25)	39 (49)	69 (35)
Severely high arched	1 (1)	10 (12)	11 (5)
Midface deficiency			
Normal	89 (74)	49 (62)	138 (69)
Mild loss of fullness	31 (26)	25 (31)	56 (28)
Substantial loss of fullness	0 (0)	6 (7)	6 (3)
Overbite			
Normal or deep bite	120 (100)	65 (81)	185 (92)
Open bite	0 (0)	15 (19)	15 (8)
Posterior bite			
Normal	118 (98)	62 (78)	180 (90)
Unilateral crossbite	2 (2)	8 (10)	10 (5)
Bilateral crossbite	0 (0)	10 (12)	10 (5)
Lip strain			
Normal	99 (82)	43 (54)	142 (71)
Mildly strained closing lips	21 (18)	24 (30)	45 (23)
Very strained closing lips	0 (0)	13 (16)	13 (6)
Lower face height			
Normal	91 (76)	45 (56)	136 (68)
Mildly excessive	28 (23)	26 (32)	54 (27)
Severely excessive	1 (1)	9 (12)	10 (5)

Values are presented as n (%). The variables are presented in order of predictor importance to clustering.

and minimal cost. The problem is that it may generate over-testing when compared to overnight oximetry or the Mallampati score.

In our analysis, we collected the evaluation of 10 independent dental specialists and then assessed their reliability. Our results showed poor to good intrarater reliability. The depicted diagnostic values were not necessarily better when additional craniofacial information and PSQ sleep questionnaire results were made available. This may indicate that relying on 3D stereophotogrammetry evaluation alone is questionable for screening OSA status among children. Additional tools that present an actual quantification of sleep-disordered breathing clinical signs and symptoms were necessary to attempt a more reliable screening approach. Even then, the performance could not be considered clinically reasonable. Indeed, it has been previously demonstrated that clinical parameters, including patient demographic information, palate position, and tonsillar size, provide limited information on the severity of OSA in children.[36]

In addition to those results, a cluster analysis was performed to understand better the role of craniofacial features in implying OSA status and probable OSA classification performance by this selected group of dental specialists. Eight craniofacial features previously associated with an increased risk of pediatric OSA were used to investigate if there was a specific craniofacial pattern in the group of children included in this study. Two clusters were identified. Cluster B presented more children with craniofacial features previously linked to pediatric OSA, specifically high arched palate and significantly increased or reversed overjet. However, the frequency of pediatric OSA patients diagnosed by nPSG or at high-risk for OSA, as suggested through the PSQ score, did not differ between clusters. Therefore, clinical judgement of risk for pediatric OSA was not improved when the craniofacial form was considered.

Specific craniofacial features defined by these clusters may have impacted how dental specialists categorized potential

Table 4—Variables used in the post hoc analysis.

	Cluster A	Cluster B	Total
Male	61 (57)	35 (48)	96 (53)
Female	46 (43)	38 (52)	84 (47)
Age (n = 196)	8.2 ± 3.5	9.8 ± 3.6	8.8 ± 3.6
BMI z-score (n = 65)	0.1 ± 1.9	1.1 ± 1.1	0.6 ± 1.6
OSA risk evaluated by PSQ (n = 200)			
At low risk	70 (58)	32 (40)	102 (51)
At high risk	50 (42)	48 (60)	98 (49)
OSA status evaluated by nPSG (n = 103)			
OSA negative	26 (53)	27 (50)	53 (52)
OSA positive	23 (47)	27 (50)	50 (48)
OSA classification based only on 3D stereophotogrammetry (n = 150)[a]			
Not likely	69 (75)	28 (49)	97 (64)
Likely	23 (25)	30 (51)	53 (36)
OSA classification based on 3D stereophotogrammetry, CFI, and PSQ (n = 144)[a]			
Not likely	57 (63)	13 (24)	70 (49)
Likely	33 (37)	41 (76)	74 (51)

Values are presented as frequency, n (%), or mean ± standard deviation. The distribution of demographic features and sleep status across groups is shown. [a]Most frequent pediatric OSA classification among all 10 clinicians. BMI = body mass index, CFI = Craniofacial Index, nPSG = nocturnal polysomnography, OSA = obstructive sleep apnea, PSQ = Pediatric Sleep Questionnaire.

pediatric OSA patients. For example, an increased overjet and a constricted palate may be linked to a compromised airway space and an increased probability of muscle collapsibility during sleep facilitating OSA.[31] The current evidence links Class II malocclusions (usually showcasing increased overjet) and constricted maxilla to pediatric OSA,[8,31,32] which may have biased the dental specialists' classification decisions. Perhaps dental clinicians overestimate the real impact of craniofacial features in pediatric OSA's complex and multifactorial entity as craniofacial morphology does not directly correlate with upper airway function.

Our findings do not support a clear categorical link between craniofacial features and OSA in children. Children with normal oropharyngeal anatomy may have OSA. This work contrasts the many studies that describe craniofacial alterations in pediatric OSA cases. Dental clinicians should not oversimplify the diagnosis and screening for pediatric OSA.

The evaluation of craniofacial features evaluation as a possible source of clinical phenotyping in children with OSA needs further probing. Many factors leading to pediatric OSA impart secondary morphologic changes in a growing patient, suggesting that some craniofacial features develop as both a cause and consequence of OSA. There is a lack of studies investigating the role of specific clinical traits in pediatric OSA, in which the available evidence is mainly focused on the nPSG sleep variables.[37–39] The dependence on the apnea-hypopnea index for diagnosis or even a communication tool to evaluate OSA severity of pediatric OSA might be challenging because this index relies only on the number of obstructive events. The reliance on this single index has been questioned due to its limited information about other OSA-relevant characteristics.[40] Information about associated comorbidities, OSA symptoms, and quality of life are still needed to establish a treatment plan or monitor treatment outcomes.[41] Nevertheless, the evaluation of facial features could help identify specific traits associated with OSA, as suggested among adult patients.[21,22]

A higher prevalence of children at high-risk for OSA has been recently reported in an orthodontic population.[42] The involvement of orthodontists and pediatric dentists in identifying OSA risk factors may improve the screening process for this disease and reduce the long-wait line for an nPSG by enhancing patients' identification at high-risk for OSA and subsequent earlier OSA diagnosis and treatment. Dental clinicians have the training and knowledge to evaluate facial features, and their involvement in the screening process, as part of a transdisciplinary group led by a sleep medicine physician, may help diagnose and treat pediatric OSA on time.

This study's overall impact would be that only patients with a clear higher risk should be referred (reduced number of false positives) to avoid further saturating the medical environment with unnecessary referrals. Over-reliance on craniofacial features as a standalone criterion should be discouraged. Much emphasis is placed on the palatal morphology of a high arched, narrow palate in children. These data suggest that these often-cited features do not consistently correlate with OSA status. Treatment is usually initiated from daytime or nighttime symptoms, and the dental practitioner has a key role in the early query of symptoms. Whether these symptoms translate to morphologic changes as the pediatric OSA patient matures is the subject of future studies.

Limitations

Not the entire sample of children had an nPSG exam that is considered a key component for a precise pediatric OSA diagnosis.

This study may have been subjected to selection bias. A convenience sample of 2 independent centers (a university orthodontic clinic and a sleep center) was included and may not reflect the general pediatric population.

In addition, the OMAHI and the cut-off of 2 events/h may have some limitations in the identification of some OSA cases. The OMAHI reports the average number of apneas and hypopneas during sleep, excluding the central respiratory events per hour in sleep.[27] However, it has been suggested that additional features, including event duration, arousal intensity, flow limitations, and obstructive hypoventilation, may also be helpful to understand pediatric OSA characteristics.[40] In future studies, these additional features should also be considered in OSA evaluation.

The definition of clusters was based on 8 features evaluated by CFI. The clinicians had access to CFI and 3D stereophotogrammetry and PSQ scores in 1 of the OSA classifications performed in this sample of children. The access to CFI information may have influenced the distribution of clinicians' judgement in clusters A and B.

The effect of the obesity and the BMI z-score was not evaluated due to the amount of missing data for a sample of the included patients. However, an increased BMI may increase the risk of children to sleep breathing disorders.[43]

The adenotonsillar size and adenotonsillectomy history were not collected in this study. Adenotonsillar hypertrophy is a risk factor for pediatric OSA and might be associated with craniofacial abnormalities.[44] Also, the presence of craniofacial anomalies, such as a smaller mandible size, were associated with residual OSA after adenotonsillectomy.[45] 3D stereophotogrammetry is a reliable method to evaluate craniofacial features among children.[46] However, the impact of different craniofacial developmental stages in assessing images obtained by 3D stereophotogrammetry has not been explored previously or in the present study.

As a time-series study, the number of children with and without craniofacial abnormalities was not matched regarding the pediatric OSA status.

Different sleep medicine physicians interpret nPSG values differently in combination with clinical exams and relevant medical history. There is no worldwide agreement on how to interpret a given set of data. Hence, the final diagnosis decision may be different when other health providers would have been involved.

No specific verbal information was provided on how to interpret the provided PSQ values. Some of the involved dental specialists may have an idea of using 8 as a cut-off. Still, others may have simply used PSQ as a continuous variable and not as a dichotomous variable.

The ethnicity of patients not evaluated in this study. The sample of this study assessed Canadian children from multiple cultures. The prevalence of bony and soft-tissue craniofacial abnormalities may vary according to ethnic groups and fat distribution. This might result in differences in OSA prevalence and severity among children.[47,48] Preterm birth history was also not considered, and it is an important risk factor.[49]

CONCLUSIONS

3D stereophotogrammetry-based facial analysis does not seem predictive for pediatric OSA screening when used alone or combined with PSQ and CFI when assessed by dental specialists interested in sleep-disordered breathing in this sample. Some craniofacial traits, more specifically significant sagittal overjet discrepancies and a high-arched palate, seem to influence participating dental specialist's classification, but these were not accurate markers of OSA.

ABBREVIATIONS

BMI, body mass index
CFI, Craniofacial Index
nPSG, nocturnal polysomnography
NPV, negative predictive value
OMAHI, obstructive–mixed apnea–hypopnea index
OSA, obstructive sleep apnea
PPV, positive predictive value
PSQ, Pediatric Sleep Questionnaire
3D, 3-dimensional

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Address correspondence to: Carlos Flores Mir, DDS, DSc, 5-528 Edmonton Clinic Health Academy, 11405 - 87 Ave NW, Edmonton, AB, T6G Canada; Tel: (780) 492-7409; Fax: (780) 492-7536; Email: cf1@ualberta.ca

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