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Craniofacial morphology and airflow in children with primary snoring

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Abstract. – OBJECTIVE: Sleep-disordered breathing (SDB) is among the most common diseases and includes a group of pathological conditions that form a severity continuum from primary snoring (PS) to obstructive sleep apnea (OSA). SDB presents a multifactorial etiology and in children, it is often linked to adenotonsillar hypertrophy, which may lead to an alteration of the breathing pattern. Therefore, several studies hinted at the existence of a correlation between SDB and the alteration of craniofacial growth. However, these studies concentrated on the most severe forms of SDB and little evidence still exists for the mildest form of SDB, namely PS. This preliminary study investigates the association between nasal airflow, measured through rhinomanometry, and cephalometric parameters in a sample of young children with PS.

PATIENTS AND METHODS: A sample of 30 children with habitual snoring aged between 5 and 8 years was selected by a SDB validated questionnaire at the Pediatric Allergology and Immunology Center of "Sapienza" University of Rome, Italy. To assess the degree of nasal obstruction, all children underwent anterior active rhinomanometry while nocturnal pulse oximetry and polysomnography were used to characterize the SDB. Cephalometric analysis was used to evaluate relevant orthodontic parameters associated to the sagittal and vertical craniofacial development and to the position of the hyoid bone.

RESULTS: We found a statistically significant association between the Frankfurt mandibular angle (FMA), which measures the total facial vertical divergence, and the severity of the airflow's obstruction ($p = 0.014$).

CONCLUSIONS: The present study supports the association between the level of nasal obstruction in children with PS and the alteration of cephalometric parameters associated with the vertical craniofacial growth, thus placing the evaluation of craniofacial parameters in the growth period in a privileged position to determine an early diagnosis of a possible insurgence of sleep disorders.

Key Words:
Craniofacial morphology, Airflow, Primary snoring, SDB, Malocclusion.

Introduction

Sleep-disordered breathing (SDB) is among the most common diseases and includes a group of pathological conditions that form a severity continuum from primary snoring (PS) to obstructive sleep apnea (OSA)[1]. Among SDB disorders, PS is not associated with gas exchange malfunctions or sleep disrupts. Conversely, OSAs are identified by repetitive partial or complete upper airway obstructions that generate ventilation disorders during sleep[2]. It should be noted that only recently snoring, which presents a prevalence of 2.5 to 34.5% of the pediatric population[3], was recognized as part of the sleep disorders. Due to its detrimental effects if not correctly diagnosed or treated, the wide scenario of SDB in children is gaining increased interest in the field of pediatric health care[4].

SDB presents a multifactorial etiology. The leading cause of SDB in children is adenotonsillar hypertrophy caused by the different growth rates of facial bones and lymphoid tissues. Other predisposing factors include craniofacial anomalies, neuromuscular diseases, obesity, and allergic rhinitis[5]. Obstructions of the upper airways due to, e.g., allergic rhinitis and/or adenotonsillar hypertrophy can induce prolonged oral respiration during critical growth periods in children and consequently initiate a sequence of events that commonly results in dental and skeletal alterations[6].

The interdependence between clinical otorhinolaryngology and orthodontics has been

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claimed by classic studies of mandibular orientation and growth in patients before and after adenoidectomy[7]. A recent paper showed some degree of normalization of the growth pattern after adenoidectomy in a group of obstructive sleep apnea patients[8]. Moreover, SDB was primarily associated with morphological features such as narrow palate and severe maxillary and mandibular crowding[9]. This close bidirectional relationship is supported nowadays by the existence of some evidence that palatal expansion can induce rhinological effects[10].

Although many authors studied associations between rhinomanometric and cephalometric parameters, the interdependence between upper airway obstruction and the craniofacial morphology can still be regarded as poorly understood and controversial. In particular, most of the researches considered only the most severe forms of SDB, characterized by apnea and/or gas exchange abnormalities, while little or no attention was devoted to milder manifestations of SDB, such as primary snoring, which conversely present a higher prevalence than OSA and can have the same risk of complications[11].

Finally, most of the studies focused on children in the adolescent age range, when the craniofacial growth process is close to its conclusion: a detailed clarification of the association between airflow and craniofacial development at the earliest possible age could lead to a multidisciplinary approach to the management and treatment of SDB, where the dental specialist would act as an early warning for SDB and then participate to its treatment pathway.

The aim of this preliminary study is, therefore, to investigate the relationship between airflow, measured through rhinomanometry, and cephalometric parameters in a sample of young children with PS.

Patients and Methods

A group of 30 children with habitual snoring (HS) aged between 5 and 8 years was selected at the Pediatric Allergology and Immunology Center of "Sapienza" University of Rome, Italy, using a questionnaire validated for SDB[12]. The questionnaire investigated sleep duration, sleep quality, and diurnal behavior alterations. A single investigator interviewed the patients and completed the questionnaires. When necessary, parents were asked to help the child without interfering with their responses. Children were considered affected by habitual snoring when the symptoms were reported three or more nights per week for at least six months.

After a screening based on the responses to the questionnaire, all subjects underwent a complete medical examination. Patients with congenital craniofacial malformations, obesity, immunosuppressive drugs intake, chronic diseases, acute illnesses, and previous orthodontic treatment were excluded. Selected subjects underwent anterior rhinoscopy, bronchodilator spirometry, and body mass index (BMI) evaluation.

The study was approved by the Ethical Committee of ''Sapienza'' University of Rome and performed with written parental informed consent. The study took place over a period of 6 months, from January to June 2014.

Anterior Active Rhinomanometry

Patients underwent Anterior Active Rhinomanometry (Sibelmed Rinospir PRO 164, Barcelona, Spain) in accordance with the International Committee on Standardization of rhinomanometry[13]. The results of rhinomanometry were considered related to nasal flows of 150 Pascal (Pa) and compared with height-dependent pediatric reference values reported in literature[14]. When the fraction of predicted values (p.v.) was in the range between 71% and 100% the rhinomanometry was considered negative (no nasal obstruction). Conversely, p.v. ≤ 70% indicated the presence of a nasal obstruction.

Following the results of the rhinomanometry, patients were divided into 5 groups according to Zapletal and Chalupova classification[15]: this classification, detailed in Table I, defines 5 levels of obstruction, going from "no obstruction" (level 1) to "very severe obstruction" (level 5).

Nocturnal Pulse Oximetry and Polysomnography

To evaluate the level of SDB, all children selected for HS underwent a nocturnal pulse oximetry and a polysomnography. These exams are here only summarized while a previous article[16] reports the technical details and fully describes how these exams were performed on the subjects of the study.

The nocturnal pulse oximetry was performed according to Brouillette et al[17] and Nixon et al[18] and measured the hemoglobin saturation (SpO2). The final result of the oximetry was classified as positive, negative, or not conclusive following

Table I. Classification of the nasal airflow obstruction in our sample according to Zapletal's criteria.

Obstruction grade	Nasal airflow fraction	Obstruction level	Number of subjects
1	71-100%	No obstruction	7 (23%)
2	57-70%	Mild	2 (7%)
3	43-56%	Moderate	7 (23%)
4	29-42%	Severe	9 (30%)
5	< 29%	Very severe	5 (17%)

the criteria detailed by Brouillette et al[19]. All children selected for HS were then admitted to the hospital and underwent an overnight polysomnography (PSG), which measured the obstructive apnea-hypopnea index (OAHI), defined as the total number of apneic and hypopneic episodes per hour of sleep.

A diagnosis of PS was given if OAHI was < 1 and SpO2 nadir was > 90%, while a diagnosis of OSA was given if OAHI was > 1. Children with OSA were then excluded from the study.

Cephalometric Analysis

All subjects underwent standard radiographical exams for orthodontics diagnosis at the Department of Oral and Maxillo-Facial Sciences, "Sapienza" University of Rome. The lateral cephalogram was taken using the natural head posture with the patient looking straight ahead, with the teeth in centric occlusion, and with the lips in light contact. All cephalometric landmarks were located and digitized by the same investigator using the OrisCeph Rx3 software and were evaluated according to Kulnis et al[20]. The same operator analyzed the cephalometric measurements for each patient using a predefined standardized approach. The measurement error, evaluated with the Dahlberg method, was always < 1 mm.

Recorded orthodontic parameters were:

1. Sagittal analysis[21] (Figure 1):
SNA (degrees): the sella-nasion-A point angle, measuring the anteroposterior relationship of the maxillary basal arch on anterior cranial base and showing the degree of maxillary prognathism.
SNB (degrees): the sella-nasion-B point angle, showing the anterior limit of the mandibular basal arch in relation to the anterior cranial base.
ANB (degrees): the A point-nasion-B point angle, defining the anteroposterior relationship of the mandible to the maxilla. This parameter was used to classify our sample according to the Angle classification (Class I: 0 ≤ ANB ≤ 4; Class II: ANB > 4; Class III: ANB < 0).
Wits appraisal (mm): the distance between the projections of the A point and the B point to the occlusal plane. This parameter allows an appraisal of the degree of anteroposterior jaw dysplasia and complements the Angle classification (Class I: -3 ≤ Wits ≤ 1; Class II: Wits > 1; Class III: Wits < -3).

2. Vertical analysis[22] (Figure 2):
FMA (degrees): the Frankfurt mandibular angle, often referred to as PFH^GoMe, is the angle between the Frankfurt horizontal plane and the mandibular plane and represents the total facial divergence. A subject is considered normodivergent when this parameter is in the range between 20 and 30.

3. Hyoid bone position[23] (Figure 3):
AH-AH' (mm): the distance between AH and its projection to the mandibular plane. With re-

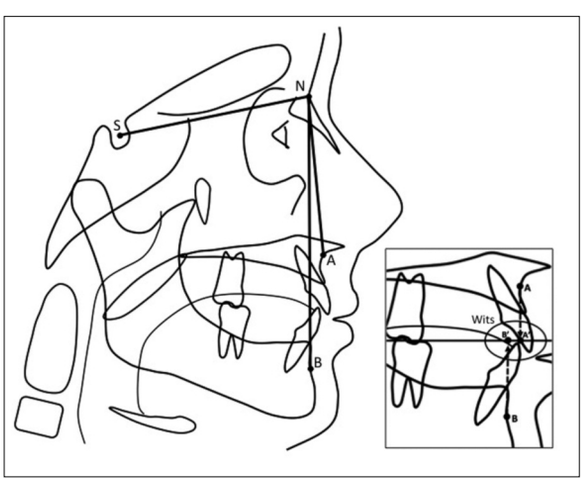

Figure 1. Cephalometric parameters – Sagittal analysis. SNA: "sella-nasion-A point" angle, measures the anteroposterior relationship of maxillary basal arch on anterior cranial base; SNB: "sella-nasion-B point" angle, measures the anterior limit of the mandibular basal arch in relation to the anterior cranial base; ANB: "A point-nasion-B point" angle, measures the anteroposterior relationship of the mandible to the maxilla; Wits appraisal (A'B'): measures the distance between the projections of points A and B to the occlusal plan.

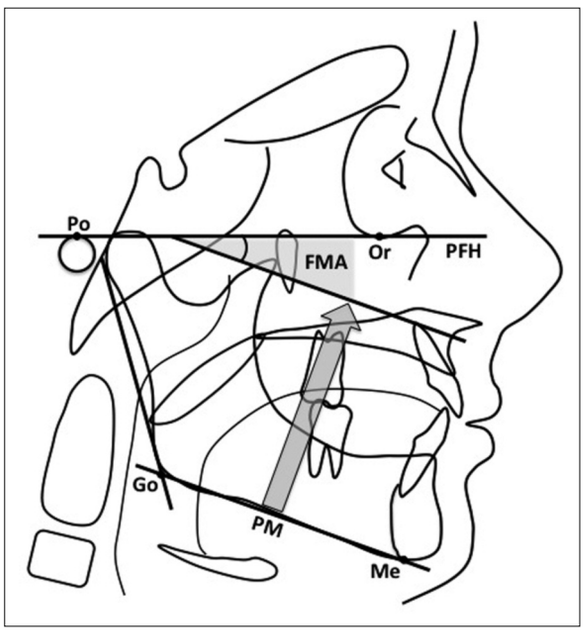

Figure 2. Cephalometric parameters – Vertical analysis. FMA: Frankfurt Mandibular Angle, measures the angle between the Frankfurt horizontal plane and the mandibular plane.

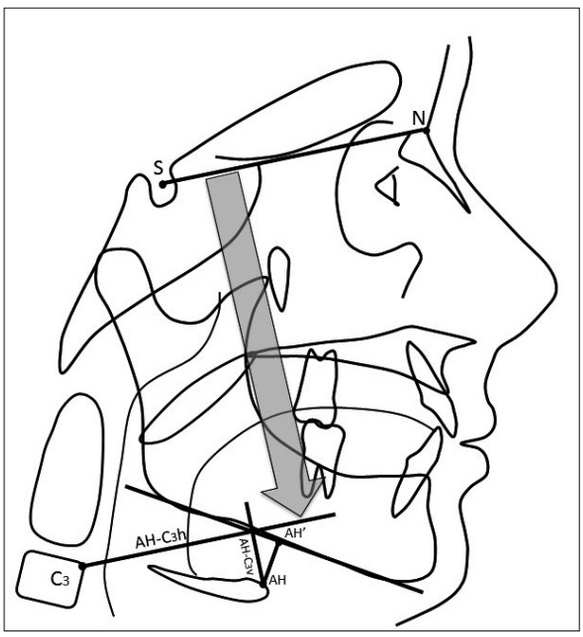

Figure 3. Cephalometric parameters – Hyoid bone analysis. AH-C3h: the horizontal distance from AH (most anterior and superior point on the body of the hyoid bone) to C3 (the third cervical vertebra) along a plane parallel to SN; AH-C3v: the vertical distance from AH to C3 along a plane perpendicular to SN; AH-AH': the distance between AH and its projection to the mandibular plane.

spect to the previous two parameters, this measure is less affected by the inclination of the cervical vertebrae.

Statistical Analysis

To perform a statistical analysis, we divided our sample into 5 groups with increasing nasal obstruction using the Zapletal and Chalupova classification of anterior active rhinomanometry. Given the small size of our sample, we could not assume a normal distribution of the data and therefore used non-parametric tests. We evaluated the homoscedasticity of the groups using the Bartlett's test and then we used the Kruskal-Wallis' test to highlight differences in the median values of each orthodontic parameter. A p-value < 0.05 was considered statistically significant.

Results

The sample included 16 female (56.7%) and 14 male (43.3%) subjects with HS. The entire sample was of Italian nationality. The age range was 5 to 8 years with a mean age of 6.73.

Table I shows the classification of the sample according to Zapletal and Chalupova criteria: nasal airflow obstruction was of grade 1 in 7 subjects (23%), of grade 2 in 2 subjects (7%), of grade 3 in 7 subjects (23%), of grade 4 in 9 subjects (30%), and of grade 5 in 5 subjects (17%).

The descriptive statistics for all cephalometric measurements are summarized in Table II.

Table II. Results of cephalometric measurements.

Cephalometric parameter	Units	Mean value	SD	Min value	Max value
SNA	Degrees	83.8	4.3	73.2	92.8
SNB	Degrees	79.2	3.8	71.2	87.4
ANB	Degrees	4.5	3.3	-3.1	13.8
Wits appraisal	mm	1.3	3.0	-3.5	9.2
FMA (PFH^GoMe)	Degrees	27.1	3.5	20.4	34.9
AH-AH'	mm	12.3	3.3	6.8	19.7

Table III. Results of the Bartlett's homoscedasticity test and of the Kruscal-Wallis' test.

Cephalometric parameter	DoF	Bartlett's χ^2	*p*	Kuskal-Wallis' χ^2	*p*
SNA	4	5.91	0.206	4.10	0.393
SNB	4	4.96	0.291	3.81	0.432
ANB	4	6.59	0.159	7.19	0.126
Wits appraisal	4	2.28	0.684	2.89	0.577
FMA (PFH^GoMe)	4	2.76	0.598	12.50	0.014
AH-AH'	4	13.52	0.009	1.81	0.771

Results of the Bartlett's test and of the Kruskal-Wallis' test are shown in Table III: the homoscedasticity hypothesis was rejected for the AH-AH' parameter ($p = 0.009$). According to the results of the Kruskal-Wallis' test, the FMA was the only orthodontic parameter that showed a statistically significant difference among the groups ($p = 0.014$). Figure 4 shows the distribution of the FMA parameter for the 5 airflow classes.

Discussion

Sleep Disordered Breathing (SDB) diagnosis in children is often difficult to obtain[4] and is frequently confused with attention deficit hyperactivity disorder. In general, there is a poor recognition of pediatric SDB in clinical practices: Blunden et al[24] found that 80% of symptomatic habitual snorers are not reported to their general medical practitioners. The failure to recognize this syndrome represents an important public health problem: Reuveni et al[25] highlighted that there is a relevant increase in health care consumption among children with sleep disorders compared with the healthy population.

Several studies[8,9,11,16,26] hinted at the existence of a correlation between the alteration of craniofacial growth features and the presence of sleep disorders. This would put the evaluation of craniofacial parameters in the growth period in a privileged position to determine an early diagnosis of a possible insurgence of sleep disorders. Following these considerations, our study investigated the correlation between several relevant cephalometric parameters and the airflow impairment, expressed according to the Zapletal and Chalupova classification, in a sample of children in the 5-8 years age range with primary snoring (PS).

We did find a statistically significant correlation between the total divergence (FMA) and the gravity of the airflow obstruction. This result suggests that the mandibular rotation plays an important role in the physiopathology of the upper airway patency.

Katyal et al[27] did not identify any cephalometric predictors in the sagittal or vertical dimensions for children at high risk for sleep disordered breathing, this in apparent disagreement with our findings. This contradiction could be explained both by the different age range of the samples (5-8 years *vs*. 8-17 years) and by the different design of the two studies: our classification is based on a direct dynamic evaluation of the airflow while the other study used a classification based on self-assessment questionnaires and cephalometric measurements of the airflow space.

Other authors[8,28] identified the vertical growth pattern of the mandible as a specific cephalometric feature in children with obstructive sleep apnea (OSA), giving an indirect support to our findings. Also, although craniofacial characteristics may predispose to OSA, some scholars[27] did not find a significant correlation between

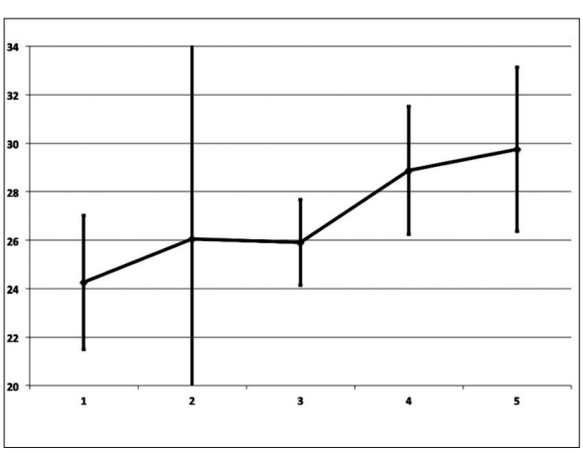

Figure 4. Mean value and 95% C.I. distribution of FMA values for the 5 airflow classes. Due to low statistics, the error bar for class 2 is not shown in full.

the long face and OSA. This can be explained by the existence of different key factors to determine the development of OSA, namely soft tissue change during growth (e.g. adenotonsillar hypertrophy) and neuro-muscular disturbances during sleep[28].

The relatively recent introduction of Cone Beam Computed tomography in orthodontics renewed the interest in upper airway characterization[29,30]. Although this technique allows a very precise transversal and volumetric airway evaluation, as of now, lateral radiography is still cheaper, less invasive, and easily accessible[31]. The drawback represented by the two-dimensional view may be minimized by using well-defined, standardized imaging and analysis techniques. Taking into account that in modern orthodontics the normal range of most cephalometric parameters is well established, we selected a set of measurements, which would be easily available to both pediatric dentists and general practitioners.

Conclusions

Cephalometric analysis can, therefore, provide to the pediatric dentist an easily accessible early warning sign of the possible presence or future development of SDB problems, suggesting the referral to an otolaryngology specialist. It would then be useful to make medical and dental practitioners aware about the importance of orthodontic issues in subjects with SDB, to identify risks at an early stage and insert the orthodontic evaluation into a multidisciplinary clinical approach to these syndromes.

Conflict of Interest

The Authors declare that there are no conflicts of interest.

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