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OBSTRUCTIVE SLEEP APNOEA AND MALOCCLUSION IN EARLY PAEDIATRIC PATIENTS: A CROSS-SECTIONAL SURVEY

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PURPOSE. To evaluate associations between obstructive sleep apnoea syndrome (OSAS) and malocclusion features in early paediatric patients.

MATERIALS AND METHODS. Children from 3 to 8 years old began a therapeutic care pathway for the diagnosis and treatment of OSAS. All subjects underwent clinical dental examination to record the following data: dental occlusion (Angle Class I, II, III); skeletal and dental malocclusion (open bite, crossbite, deep bite, overjet) and narrow palate. A polysomnographic examination was conducted to obtain a definitive diagnosis of OSAS, and two groups were identified: OSAS+ (Group 1) and OSAS- (Group 2). Chi-squared testing was performed to compare variables between the two groups. Odds ratio testing was performed to explore statistically significant values.

RESULTS. Two hundred and nine children were assessed, 92 of which were classified as OSAS+ and 117 OSAS-. No statistically significant differences were found between groups for Angle Class I, II or III, open bite, deep bite, or greater overjet (P >0.05). Statistically significant differences were found between groups for crossbite, pure and mixed, (P = 0.009; OR 2.54; 95% CI 1.25-5.16), and narrow palate (P = 0.001; OR 2.55; 95%CI 1.45-4.49)

CONCLUSIONS. Certain malocclusions, specifically crossbite and narrow palate, could be associated with obstructive sleep apnoea syndrome in children.

CONFLICT OF INTEREST STATEMENT
The authors have no conflicts of interest to disclose.

INTRODUCTION

Obstructive sleep apnoea syndrome (OSAS) is a respiratory disorder characterized by repeated episodes of complete or partial obstruction of the upper airways during sleep. It can affect both children and adults[1], in whom the obstruction leads to episodes of apnoea or hypopnea, respectively defined as an interruption of breathing for a period ranging from 10 seconds to a maximum of 180 seconds, and a reduction in breathing[2,3].

According to the America Academy of Sleep Medicine (AASM), the diagnosis of OSAS is to be based on both daytime and nocturnal symptoms, as well as a well-documented full-night polysomnography.

The apnoea-hypopnea index (AHI) is used to define the severity of OSAS by calculating the sum of the apnoea and hypopnea episodes per hour of sleep. OSAS can be thereby be classified as mild (1 to 4 episodes per hour), moderate (5 to 10 episodes per hour) or severe (more than 10 episodes per hour)[4].

Several studies have reported that almost 50% of children present at least one or more of the symptoms related to sleep-related breathing problems[5,6]. OSAS has a prevalence of 1% to 5% in individuals aged 2 to 12 years[7], with a peak incidence in the age group 2 to 8 years[8-10].

Several risk factors that can facilitate the onset of OSAS in the paediatric age have been reported[11].

Among these, the best documented risk factors are the following.

- *Obesity*. This mainly impacts OSAS onset through two supposed mechanisms: (i) fat at the pharyngeal soft tissue level decreases the lumen's diameter and speeds up the collapse of the structures themselves, and (ii) these patients' respiratory performance is greatly worsened by increasing fat accumulation in their thoracic and abdominal walls[12,13].
- *Adenoid and/or tonsil hypertrophy*. The lymphoid tissue of the Waldeyer ring is more developed at an age of between 3 and 6 years, in line with the peak incidence of OSAS[14]. The retro-palatal region has the smallest cross-sectional area, and is consequently the most frequent source of obstruction due to oropharyngeal constriction and lateral collapse. Children with Grade IV tonsils (kissing tonsils) are particularly prone to developing sleep difficulties[11-15].
- *Allergic rhinitis*. This can be responsible for OSAS since nasal congestion, secondary to inflammation of the nasal mucosa, induces an increase in the resistance of the upper airways and can cause oral breathing, sleep disturbances and fatigue[16].
- Craniofacial abnormalities and genetics. Upper airway obstruction syndrome can also be associated with craniofacial anomalies. The retro-palatal region, which is the most common source of obstruction in juvenile patients, could be a result of changes in the mandible's size, position, and geometry, as well as changes in the size of the tongue[17,18].

Some genetic problems, including as Down, Prader-Willi and Beckwith-Wiedemann syndromes, have been linked to OSAS, but the pathophysiology of paediatric OSAS is still unclear[19-21]. Nevertheless, craniofacial abnormalities in terms of malocclusions have been suggested to play a role[22]. Most of the evidence in the literature points to a major association between OSAS and Angle second class, as well as increased overjet, crossbite and narrow palate[3,22,23].

The purpose of this cross-sectional survey was to assess possible correlations between obstructive sleep apnoea syndrome and malocclusion in early paediatric patients. This study is reported in line with STROBE guidelines (https://www.strobe-statement.org/checklists/).

MATERIALS AND METHODS

Children with suspected diagnosis of OSAS were assessed at the University Hospital of Sassari, Italy. Their parents were duly informed about the nature of the study and signed a dedicated informed consent form. Ethical review and approval were waived for this study due to its retrospective observational design, and no mandatory ethics approval is required by Italian law (GU No. 76 31/Mar/2008).

Inclusion criteria were:

- subjects between the ages of 3 and 8.

Exclusion criteria were:

- subjects who presented other comorbidities (for example, Down syndrome, macroglossia);
- subjects who had undergone adenotonsillectomy (AT);
- subjects undergoing orthodontic/myofunctional therapy.

Clinical assessment steps

The paediatric diagnostic-therapeutic care pathway involved the collaboration of different healthcare professionals, including paediatricians, dentists and otolaryngologists. The purpose of this pathway was to facilitate the diagnosis of OSAS, find the most appropriate treatment for these children to reduce healthcare costs. The pathway is divided into three phases, the first of which involved children with suspected OSAS being assessed by paediatricians (R.A. and C.L.), who compiled a clinical sleep record[24] (CSR) to provide support for a diagnosis of OSAS or not. In the second phase, the children were clinically examined by an otolaryngologist, and in the third a specialist clinical dental assessment was carried out by dentists (A.L and G.S).

After completing the previously described steps, the children underwent a polysomnographic examination to receive a definitive diagnosis of OSAS+ or OSAS-. Then, the apnoea/hypopnea index (AHI) was calculated, classifying OSAS+ patients as either mild OSAS (AHI = 1 to 4 episodes for sleeping hour), moderate (AHI = 5 to 10 episodes for sleeping hour) or severe (AHI > 10 episodes for sleeping hour).

Patients' characteristics

The following conditions were analysed in an unblinded fashion.

- *Dental occlusion*: the occlusal relationship between maxillary and mandibular teeth was assessed according to the modified Angle's classification for primary dentition[25]. Under this system, Class I is diagnosed when the mesiobuccal cusp of the primary maxillary second molar occludes with the mesiobuccal groove of the primary mandibular second molar; Class II when the mesiobuccal cusp of the primary maxillary second molar occludes with the interdental space between the primary mandibular first and second molars; and Class III when the mesiobuccal cusp of the primary maxillary second molar occludes with the distobuccal groove or distal surface of the primary mandibular second molar.
- *Skeletal and/or dental malocclusions*. Among these were evaluated:
  - open bite: no vertical overlap or contact between the anterior incisors;
  - crossbite, pure or mixed: misalignment of dentition wherein the upper teeth fit inside the lower teeth;
  - deep bite: the upper front teeth excessively overlap (more than 4 mm) the bottom front teeth when the back teeth are closed;
  - greater overjet: the horizontal (anterior–posterior) overlap the maxillary central incisions over the mandibular central incisors by more than 2 mm.
- *Narrow palate*: assessed clinically as when the morphology of the palate is narrower than normal conditions.

Statistical analysis

The statistical analysis was carried out by a dental clinician expert in biostatistics (D.M.) using an open-source calculation software called Jamovi (Version 2.3), based on R-type language. To determine statistical differences between groups, the chi-squared test was applied, assuming the independence between variables as the null hypothesis (H0), with dependent variables as the alternative hypothesis (H1). Where a statistically significant P value was found, odds ratio with 95% confidence interval is reported. All statistical analyses were performed with a significance value set at 95% ($\alpha = 0.05$).

RESULTS

From March 2019 to February 2022, 209 children were assessed, 70 females and 139 males with median age 4±0.7 years (2 to 8 years). After polysomnography, 92 subjects were classified as OSAS+ (Group 1: 35 females and 57 males of median age 3.8±0.8 years) and 117 subjects as OSAS- (Group 2: 35 females and 82 males of median age 4.0±0.6 years). Among Group 1, 36 children (17.2%) displayed an AHI >10 (severe OSAS), 40 children (19.1%) an AHI of 5-10, (moderate OSAS), and 16 children (7.7%) mild OSAS.

The occlusal characteristics of both groups are shown in **TABLES 1 AND 2**, respectively. No statistically significant differences between groups were found in terms of age (P = 0.40; **TABLE 3**) or sex (P = 0.21; **TABLE 4**).

Likewise, no statistically significant differences were found between groups for Angle Class I, II or III (P = 0.995; **TABLE 5**), greater overjet (P = 0.72; **TABLE 6**), open bite (P = 0.85; **TABLE 7**) or deep bite (P = 0.15; **TABLE 8**). However, statistically significant differences between groups were found for crossbite, both pure and mixed (P = 0.009; OR 2.54; 95% CI 1.25-5.16; **TABLE 9**) and narrow palate (P = 0.001; OR 2.55; 95%CI 1.45-4.49; **TABLE 10**).

TABLE 1 OCCLUSAL CHARACTERISTICS OF OSAS+ SUBJECTS

OSAS+	Number of subjects N = 92	
Dental occlusion		
I Class	58	63.04%
II Class	25	27.17%
III Class	9	9.79%
Bite characteristics		
Open bite	16	17.39%
Open/crossbite bilateral	2	2.17%
Open/crossbite monolateral	3	3.26%
Crossbite bilateral	17	18.48%
Crossbite monolateral	1	1.09%
Correct bite	27	29.35%
Deep bite	19	20.65%
Deep/crossbite	2	2.17%
Edge-edge	5	5.43%
Overjet		
Underbite	10	10.87%
Correct overjet	54	58.70%
Higher overjet	28	30.43 %
Palate		
Narrow palate	61	66.30%
Normal palate	31	33.70%

TABLE 2 OCCLUSAL CHARACTERISTICS OF OSAS- SUBJECTS

OSAS-	Number of subjects N = 117	
Dental occlusion		
I Class	74	63.25%
II Class	32	27.35%
III Class	11	9.40%
Bite characteristics		
Open bite	14	11.97%
Open/crossbite bilateral	1	0.85%
Open/crossbite monolateral	1	0.85%
Crossbite bilateral	10	8.55%
Crossbite monolateral	2	1.71%
Correct bite	50	42.74%
Deep bite	36	30.77%
Deep/crossbite	1	0.85%
Edge–edge	2	1.71%
Overjet		
Underbite	11	9.40%
Correct overjet	73	62.39%
Higher overjet	33	28.21%
Palate		
Narrow palate	51	43.59%
Normal palate	66	56.41%

TABLE 3 ANALYSIS OF AGES IN OSAS+/-

	OSAS+	OSAS-
Age (Mean±SD)	3.8±0.8	4.0±0.6
P value	0.400	

TABLE 4 ANALYSIS OF SEX IN OSAS +/-

SEX	OSAS+	OSAS-	Total
Female	35	35	70
Male	57	82	139
Total	92	117	209
P value	0.216		

TABLE 5 ANALYSIS OF ANGLE CLASS IN OSAS+/OSAS-

Angle Class	OSAS+	OSAS-	Total
Angle I Class	58	74	132
Angle II Class	25	32	57
Angle IIII Class	9	11	20
Total	92	117	209
P value	0.995		

TABLE 6 ANALYSIS OF GREATER OVERJET IN OSAS+/ OSAS-

Greater overjet	OSAS+	OSAS-	Total
No	64	84	148
Yes	28	33	61
Total	92	117	209
P value	0.724		

TABLE 7 ANALYSIS OF OPEN BITE IN OSAS+/OSAS-

OPEN BITE	OSAS+	OSAS-	TOTAL
No	71	101	172
Yes	21	16	37
Total	92	117	209
P value	0.853		

TABLE 8 ANALYSIS OF DEEP BITE IN OSAS+/OSAS-

DEEP BITE	OSAS+	OSAS-	TOTAL
No	71	80	151
Yes	21	37	58
Total	92	117	209
P value	0.158		

TABLE 9 ANALYSIS OF CROSSBITE IN OSAS+/OSAS-

CROSSBITE	OSAS+	OSAS-	TOTAL
No	67	102	169
Yes	25	15	40
Total	92	117	209
P value	0.009		

TABLE 10 ANALYSIS OF NARROW PALATE IN OSAS+/OSAS-

NARROW PALATE	OSAS+	OSAS-	TOTAL
No	31	66	97
Yes	61	51	112
Total	92	117	209
P value	0.001		

DISCUSSION

Paediatric OSAS represents a pathological condition that requires a multidisciplinary approach, both in terms of diagnosis but also treatment[22]. The purpose of this study was to assess for a possible link between malocclusion and OSAS, which has been postulated in the literature but not yet proven conclusively[22].

In our study we found a statistically significant difference between OSAS+ and OSAS- groups for crossbite and narrow palate, but no statistically significant difference between groups for Angle Class I, II and III, greater overjet, deep bite or open bite. This confirms findings by several authors. For instance, Pisacane et al.[22], in a retrospective cross-sectional survey, found no relationship between sagittal malocclusion (Angle Class) and OSAS, as did Carvalho et al.[26], in their pilot study. Like us, these latter authors also found no correlation with greater overjet, but that OSAS was associated with crossbite. Other authors, including Hultcrantz et al.[27] in their retrospective cross-sectional survey, and Hansen et al.[28] and Katyal et al.[29] in systematic reviews, indicated the same malocclusions - crossbite and narrow palate - as risk factors for the onset of OSAS.

A possible anatomical explanation for these findings is that the palate forms the floor of the nasal cavities, and consequently any condition that alters the physiological morphology of the palate will have an effect on the nasal cavities. The presence of a high palatine vault or a contracted upper jaw, resulting in the establishment of a crossbite, shrinks the space at the base of the nasal cavities, reducing air flow and increasing resistance[29]. Our study seems to corroborate the evidence in this regard presented to date.

The main limitation of this survey is that the patient characteristics were assessed only clinically, without any instrumental or radiological evaluation, although this was to avoid exposing children to unnecessary x-ray exposure. In particular, the narrow versus normal palate classification was assessed without using any objective, reproducible numerical values. These findings need to be confirmed by prospective clinical studies. Nevertheless, the data obtained lend weight to the idea that specific interceptive orthodontic treatments could be useful in selected cases of OSAS[30-32].

CONCLUSIONS

Despite the limitations of the study, our findings support the hypothesis that crossbite and narrow palate could be risk factors for early OSAS onset.

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