

Relationship between Oral Parafunctions and Signs and Symptoms of Craniomandibular Dysfunction in Children with Cleft Lip and Palate

Relação Entre Parafunções Oraís e Sinais e Sintomas de Disfunção Craniomandibular em Crianças com Fissura Labiopalatina

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Abstract

The aim of this study was to evaluate the association of oral parafunctions with the presence of signs and symptoms of craniomandibular disorder in children with cleft lip and palate (CLP). A cross-sectional study was performed in which one hundred and eighty-eight children with CLP and absence of associated syndromes or malformations were investigated. During clinical examination, the presence of wear facets and dental occlusion were observed and palpation was performed in the temporomandibular joint region (TMJ) and orofacial musculature. The children and their guardians were interviewed regarding the presence of oral parafunctional habits (bruxism, the act of biting the lips and/or cheeks, nail biting, sucking of fingers and pacifiers) and signs and symptoms of craniomandibular dysfunction (headache, ear and/or neckache, tiredness or crack in the TMJ during mouth opening and chewing movements). The data were analyzed by means of absolute and relative frequency measurements, and bivariate analysis using Pearson's chi-square test and likelihood ratio test were performed, considering the significance level of 5%. A higher prevalence of cleft lip and palate type (76.0%) and the presence of malocclusion (61.7%) was observed, highlighting the anterior crossbite (58.7%) and midline deviation (48.9%). There was a higher occurrence of teeth grinding (40.4%), onychophagy (28.7%) and biting lips and/or cheeks (28.7%). Onychophagia was associated with headache ($p < 0.05$) and with pain or cracking during mouth opening ($p < 0.05$). It was concluded that parafunctional oral habits may be associated with signs and symptoms of craniomandibular disorder in children with cleft lip and palate.

Keywords: Craniomandibular Disorders. Nail Biting. Bruxism. Maxillofacial Abnormalities.

Resumo

O objetivo do estudo foi avaliar a associação de parafunções orais com sinais e sintomas da disfunção craniomandibular em crianças com fissuras labiopalatinas (FLP). Um estudo transversal foi realizado, no qual cento e oitenta e oito crianças com FLP e ausência de síndromes ou malformações associadas, foram investigadas. Durante exame clínico, a presença de facetas de desgaste e a oclusão dentária foram observadas e foi realizada a palpação na região da articulação temporomandibular (ATM) e musculatura orofacial. As crianças e seus responsáveis foram entrevistados com relação à presença de hábitos orais parafuncionais (bruxismo, ato de morder os lábios e/ou bochechas, onicofagia, sucção de dedos e de chupeta) e sinais e sintomas de disfunção craniomandibular (cefaléia, ouvido, dor, cansaço ou estalo na ATM durante movimentos de abertura bucal e de mastigação). Os dados foram analisados por meio de medidas de frequência absoluta e relativa e a análise bivariada foi realizada por meio dos testes do Qui-quadrado de Pearson (χ^2) e a Razão de verossimilhança, considerando o nível de significância de 5%. Uma maior prevalência das fissuras envolvendo lábio e palato (76,0%) e presença de maloclusão (61,7%) foi observada, com destaque para mordida cruzada anterior (58,7%) e desvio de linha média (48,9%). Houve uma maior ocorrência do ato de ranger os dentes (40,4%), da onicofagia (28,7%) e do hábito de morder lábios e/ou bochechas (28,7%). A onicofagia foi associada à cefaléia ($p < 0,05$) e à dor ou estalo durante abertura bucal ($p < 0,05$). Conclui-se que hábitos orais parafuncionais podem estar associados a presença de sinais e sintomas da disfunção craniomandibular em crianças com FLP.

Palavras-chave: Transtornos Craniomandibulares. Hábito de Roer Unhas. Bruxismo. Anormalidades Maxilofaciais.

1 Introduction

Among the congenital craniofacial anomalies, cleft lip and palate (CLP) is one of the most frequent¹, with multifactorial etiology^{2,3}. Epidemiological data vary according to region and country^{5,6}, highlighting a higher prevalence in Caucasian individuals when compared to Asians and Africans⁶. It is estimated that cleft lip with or without involvement of the palate is 1: 1000 births and cleft palate is 1: 2500⁷. Frequently, CLP affects males in a ratio of 2: 1 to females^{5,6}, however when it comes to isolated cleft palates there is an inversion, with a

higher prevalence in females^{5,7}. In Brazil, the prevalence is 4.85 per 10,000 live births, varying according to the country's regions⁸.

Individuals with CLP, especially those in which anatomical defects involve the alveolar ridge, have dental alterations, such as agenesis of lateral incisors, supernumerary teeth, dental crowding, abnormalities in the tooth shape, hypodontia, and changes in the dental eruption chronology⁹⁻¹⁴. In addition, they have occlusal disorders resulting from maxillary atresia, such as posterior crossbite and the class III profile¹⁵. Due to the morphological changes, individuals with CLP may face

changes in the functional bases of the stomatognathic system, such as sucking, swallowing, chewing, speech articulation¹⁶⁻¹⁸ and breathing^{17,19}.

The presence of FLP seems to affect individuals emotionally²⁰, since dissatisfaction with a self-image, difficulty in relationships among people, as well as reduced self-confidence and well-being in different age groups were observed²¹, which could negatively affect the quality of life of CLP population^{22,23}. In this context, children who are shy, anxious and more tense due to the presence of face malformation become strong candidates for the practice of parafunctional habits²⁴⁻²⁷.

Parafunction is a damage acquired by constant repetition, becoming harmful when it exceeds the tolerance limit, that is, when the repetition of the movement and the duration of the force is prolonged during the child's growth and craniofacial development²⁸. Oral parafunctions include bruxism (teeth grinding), dental clenching, the act of biting objects, lips and/or cheeks, onychophagia (biting nails) and non-nutritive sucking (pacifier and finger sucking)^{26,29,30}. The impact of oral parafunction on the stomatognathic system is significant, since there is an association between oral functions altered by the pathology itself and the consequences of the habit¹⁶⁻¹⁹. The deleterious effects of oral parafunctions are characterized by mouth breathing accompanied by nocturnal sialorrhea²⁶, sleep apnea¹⁹, changes in the usual tongue posture and facial muscle tone²⁶, changes in dental positioning and dental occlusion^{17,28}.

Oral parafunctions, emotional factors (anxiety and stress)^{27,31}, and temporomandibular joint (TMJ) disorders³² are factors that, individually or together, can trigger the development of craniomandibular disorder (CMD). CMD is a term used to represent a set of signs and symptoms that affect the facial and masticatory muscles, the temporomandibular joint and other associated structures^{33,34}. Although this disorder is still little reported in the childhood, a limitation during mouth opening and the presence of sounds at the TMJ, headache, tenderness during palpation of the TMJ and chewing muscles were observed^{26,29,30,35-37}, which can be aggravated with the child's development^{28,35}.

Based on little evidence regarding oral parafunctions and CMD in children with CLP, the aim of this study was to evaluate the association among oral parafunctions and signs and symptoms of craniomandibular disorder in children with cleft lip and palate. The null tested hypothesis was that there is no relationship between the presence of oral parafunctional habits and signs and symptoms of craniomandibular disorder in children with CLP.

2 Material and Methods

2.1 Ethical aspects

This study was approved by the Research Ethics Committee of University of Cuiabá-UNIC (CAAE: 13304513.4.0000.5165). Children and parents/guardians

signed the Assent and Informed Consent Forms, respectively.

2.2 Selection of the study population

Children aged between 5 and 12 years who attended the Cleft Lip and Palate Service at General Hospital of Cuiabá, Cuiabá, Mato Grosso, Brazil, between 2014 and 2018, were invited to participate in this study. The sample calculation was carried out on the total eligible population (203 individuals), considering a significance level of 5%, statistical power of 0.80 and minimum confidence interval from 0.05 to 0.10 (National Statistical Service & Sample Size Calculator Definitions - NSS), requiring at least 133 participants.

The inclusion criteria of individuals were children with cleft lip, cleft lip and palate or isolated cleft palate, both sexes and who underwent primary cheiloplasty and palatoplasty surgeries. Individuals with other malformations or syndromes associated, systemic diseases or in chronic use of psychotropics were excluded from the study population.

2.3 Interview and clinical examination

The children and their guardians were interviewed regarding the presence of oral parafunctional habits (bruxism, biting the lips and/or cheeks, onychophagia, digital and pacifier sucking) and signs and symptoms of craniomandibular disorder (headache, ear and/or neckache, TMJ tiredness or popping during mouth opening and chewing movements).

The clinical examination was performed by two previously trained professionals. The intra-examiner and inter-examiner calibration was performed using Kappa concordance analysis. For this purpose, 10 children were evaluated twice by the two examiners, with an interval of 7 days. During the extraoral examination, palpation was performed at the TMJ region and in orofacial muscles (masseter, temporal, cervical and sternocleidomastoid muscles), and the pattern of breathing and swallowing was observed. The intraoral examination was performed using a clinical mirror, and the presence of wear facets, dental occlusion and midline were evaluated.

2.4 Statistical analysis

The data were analyzed by means of absolute and relative frequency measurements. The association among the variables was performed using Pearson's chi-square test (χ^2) and the Likelihood Ratio Test, considering the significance level of 5%.

3 Results and Discussion

Of the 202 individuals invited to participate, 14 were excluded for having associated syndromes or for refusing to participate in the study. Thus, 188 children aged 5 to 11 years were included in the final population. The Kappa coefficients obtained from the intra and inter-examiner calibration were 0.88 and 0.84, respectively.

The characterization of the population regarding sex, age,

type of cleft, occlusion and midline is shown in Table 1. A higher prevalence of cleft lip and palate type (76.0%) and the presence of malocclusion (61.7%) were observed, highlighting the anterior crossbite (58.7%) and midline deviation (48.9%).

Table 1 - Description of the study population (N=188)

Characteristic	N (%)
Gender	
Female	78 (41.49)
Male	110 (58.51)
Age (years)	
5	31 (16.49)
6	46 (24.47)
7	31 (16.49)
8	29 (15.43)
9	23 (12.23)
10	15 (7.98)
11	13 (6.91)
Cleft type	
Lip	30 (15.96)
Palate	42 (22.34)
Lip and Palate	116 (61.70)
Occlusion	
Normocclusion	45 (23.94)
Malocclusion	143 (76.06)
Anterior crossbite	84 (58.74)
Unilateral posterior crossbite	23 (16.08)
Excessive horizontal overhead	15 (10.49)
Bilateral posterior crossbite	10 (6.99)
Excessive vertical overhead	8 (5.59)
Anterior open bite	3 (2.10)
Midline deviation	
Presence	92 (48.94)
Absence	96 (51.06)

Source: Research data.

Regarding the presence of parafunctional oral habits, a

higher occurrence of bruxism, onychophagia and biting lips and/or cheeks was observed (Table 2). Signs and symptoms of CMD were observed in CLP children, highlighting the headache, earache and pain or cracking during mouth opening (Table 3).

Table 2 - Parafunctional oral habits of the study population (N=188)

Parafunctional Habits	Presence N (%)	Absence N (%)
Bruxism	76 (40.43)	112 (59.57)
Biting lips and/or cheeks	54 (28.72)	134 (71.28)
Onychophagy	54 (28.72)	134 (71.28)
Digital suction	12 (6.38)	176 (93.62)
Pacifier sucking	3 (1.60)	185 (98.40)

Source: Research data.

Table 3 - Description of signs and symptoms of craniomandibular disorder (CMD) in children with cleft lip and palate (N=188)

Signs and Symptoms of CMD	Presence N (%)	Absence N (%)
Headache	73 (38.83)	115 (61.17)
Earache	46 (24.47)	142 (75.53)
Neckache	9 (4.79)	179 (95.21)
Tiredness during mouth opening	10 (5.32)	178 (94.68)
Pain or tiredness while chewing	9 (4.79)	179 (95.21)
Pain or crackling during mouth opening	26 (13.83)	162 (86.17)

Source: Research data.

When assessing the association of signs and symptoms of CMD with the presence of parafunctional oral habits, a significant result was observed for onychophagia and headache ($p < 0.05$) and, onychophagia and pain or cracking during mouth opening ($p < 0.05$; Table 4), rejecting the null hypothesis tested in this study.

Table 4 - Association between parafunctional habits and signs and symptoms of CMD in children with cleft lip and palate (Chi-square test; $p < 0.05$)

	Headache			Earache			Neckache			Pain or crackling during mouth opening			Pain or tiredness while chewing		
	Yes	No	P value	Yes	No	P value	Yes	No	P value	Yes	No	P value	Yes	No	P value
Bruxism															
Presence	26	50	0.284	18	58	0.837	4	72	1,000 FE*	10	66	0.826	3	73	0.741 FE
Absence	47	65		28	84		5	107		16	96		6	106	
Biting the lips and/or cheeks															
Presence	24	30	0.316	15	39	0.503	3	51	0.718	9	45	0.474	5	49	0.123 FE
Absence	49	85		31	103		6	128		17	117		4	130	
Onychophagy															
Presence	27	27	0.046	16	38	0.296	5	49	0.123	12	42	0.034	3	51	0.718 FE
Absence	46	88		30	104		4	130		14	120		6	128	
Digital suction															
Presence	5	7	0.835	2	10	0.733 FE	0	12	0.508 LR	1	11	1.000 FE	1	11	0.455 FE
Absence	68	108		44	132		9	167		25	151		8	168	
Pacifier sucking															
Presence	3	0	0.057 LR**	0	3	0.577 LR	0	3	1.000 LR	0	3	0.689 LR	0	3	1.000 LR
Absence	70	115		46	139		9	176		26	159		9	173	

*FE: Fisher's exact test; **LR: Likelihood-ratio test

Source: Research data.

The cleft lip and palate type of cleft showed greater occurrence in the investigated population (61.7%), agreeing with previous investigations^{5,38-41}. Malocclusion was present in 76% of the subjects, regardless of the type of cleft, being in accordance with previous studies^{14,40-44}.

Parafunctional oral habits are frequently found in children since they are the result of persistent physiological habits^{28,45}. In this investigation, a higher prevalence of bruxism, onychophagia and the biting lips and/or cheeks was observed, and an absence of pacifier use (98.4%), which is in accordance with previous studies with children with CLP^{18,40} and without CLP^{26,45}. In previous case-control studies, the highest occurrence of oral parafunctional, such as onychophagy, digital suction, bringing objects in the mouth, lip and/or cheek suction, inappropriate tongue movements, snoring, mouth breathing and bruxism were observed in children with CLP when compared to children without the deformity^{16,18}.

The low prevalence of pacifier use in children with CLP may occur due to difficulty performing suction because of the presence of the anatomical defect, that can impair the effective intraoral pressure^{4,18}. Also, young children are encouraged to interrupt pacifier sucking habit, after the repair surgeries (cheiloplasty and palatoplasty) to prevent trauma, fistula and infection^{16,18}.

Nails biting and grinding teeth are associated with the individual's emotional state^{25,46}, which may cause functional disorders and also negative psychosocial experiences^{23,46}. Thus, in this study, the occurrence of these habits could be explained due to the fact that children with CLP generally have a significant emotional burden, due to the numerous medical consultations, surgeries, impairment of aesthetics and difficulty in the social relationship⁴⁷.

The most observed signs and symptoms of CMD, in this study, were headache, earache, and pain or crackling during mouth opening movement, corroborating with previous studies with children and adolescents without the malformation^{30,35,45,48-51}. A previous study observed, in children with CLP, muscle and TMJ sensitivity, TMJ noise and limited mouth opening⁵².

For the diagnosis of CMD in children, pain, joint sound and limited mouth opening are considered, which may appear combined or not⁵³. Alone, signs and symptoms do not represent the development of the disease, although headache appears to be a risk factor for CMD in children^{50,54,55}, which reinforces the investigation of the other factors^{34,50,53}, as well as the presence of oral parafunction, whose deleterious effect depends on the frequency, intensity and duration of the habit⁵⁶, inadequate posture, psychosomatic symptoms (anxiety and increased level of stress)^{32,50}, trauma to the head and neck and hereditary pattern⁵³. When confirmed the presence of CMD, the treatment should be discussed by the pediatric dentist and the TMD specialist for the prevention and control of future deleterious effects⁵³, since there is an increase in the manifestations of the syndrome, which can occur with age and

pubertal status^{28,49-51}.

In this investigation, the relationship between nail biting and headache, and nail biting and pain or cracking during mouth opening was observed, being in agreement with previous findings^{29,30,37,45,48,50,56-58}. During nail biting habit, there is a greater functional demand on the facial and TMJ muscles that act differently than what occurs during chewing, resulting in trauma to the masticatory system^{29,59}. The long-term onychophagy may reduce the joint space and compression of articular disc, resulting in pain and noise during mouth opening^{48,56}. The severity of nail biting habit may increase with age, since it is related to increased anxiety and stress²⁵, which have already been showed in children with CLP⁴⁶.

There is a limitation of this study due to its cross-sectional design, since it is not possible to affirm that the presence of cleft lip and palate is determinant for the development of parafunctional oral habits or the presence of signs and symptoms of CMD. Thus, based on the limitation of the cross-sectional study and the lack of scientific evidence on the diagnosis of CMD in children with CLP, case-control and longitudinal studies are recommended to elucidate the gap.

4 Conclusion

Parafunctional oral habits may be associated with signs and symptoms of craniomandibular disorder in children with cleft lip and palate. Thus, the early diagnosis and interruption of oral parafunctions are important to prevent changes in oral functions and possible development of craniomandibular dysfunction.

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