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COMPARISON OF PERIODONTAL PARAMETERS IN INDIVIDUALS WITH SYNDROMIC CRANIOSYNOSTOSIS

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ABSTRACT

Craniosynostosis syndromes are characterized by premature closure of one or more cranial sutures, associated with other malformations, the most frequent of which are the Crouzon and Apert syndromes. Few studies in the literature have addressed the oral health of these individuals. The purpose of this study was to compare the periodontal status of individuals with Apert, Crouzon, Pfeiffer and Saethre-Chotzen syndromes before toothbrushing and compare the efficiency of plaque removal before and after mechanical toothbrushing. The probing depth, plaque index (according to Löe and O'Leary), clinical attachment level, gingival index (according to Silness and Löe) and amount of keratinized mucosa were evaluated before toothbrushing, and the O'Leary plaque index was assessed before and immediately after toothbrushing, on the same day, in 27 individuals aged 11 to 36 years. There was statistically significant difference in the mean probing depth and clinical attachment level among regions (p=0.00; p=0.01, respectively). The gingival index did not reveal statistically significant differences. With regard to the plaque index, the left region exhibited higher plaque index values than the right and anterior regions. No significant results were found in the analysis of keratinized mucosa. Comparison of the O'Leary plaque index before and after toothbrushing revealed statistically significant difference for all syndromes except for the Pfeiffer syndrome ($p<0.05$). In conclusion, there was no difference in the periodontal status among individuals with syndromic craniosynostosis. The posterior region was more affected than the anterior region as to the presence of plaque, loss of insertion and probing depth. Individuals with Pfeiffer syndrome exhibited greater toothbrushing efficiency than individuals with the other craniosynostosis syndromes.

Key words: Craniosynostosis. Craniostenosis. Periodontal index. Epidemiology.

INTRODUCTION

The term craniosynostosis or craniostenosis is characterized by the premature closure of one or more cranial sutures[29]. This closure causes a reduction in bone synthesis, leading to craniofacial deformities. The craniosynostoses may be classified according to the number of sutures affected (simple or multiple), cause (primary or of unknown cause and secondary or of known cause), or according to the association with other malformations (non-syndromic and syndromic). Simple, primary and non-syndromic craniosynostoses are the most common[12].

The appearance of craniosynostosis may be related with both environmental and genetic factors. During the last decade, it has been observed that mutations in four genes may lead to the occurrence of syndromic craniosynostoses, such as Apert, Pfeiffer, Crouzon and Saethre-Chotzen syndromes. The most frequent craniosynostosis syndromes are the Crouzon and Apert syndromes, which are usually hereditary; with rare exceptions, the diagnosis is established at birth[13]. Apert, Crouzon, Pfeiffer and Saethre-Chotzen syndromes exhibit similar systemic and oral changes in addition to some changes inherent to each syndrome (Figure 1).

One of the few studies in the literature addressing the oral health of these individuals was conducted by Mustafa, et al.[27] (2001), who examined 57 children with craniosynostosis (11 Apert, 21 Crouzon, 5 Pfeiffer, 3 Saethre-Chotzen and 17 non-syndromic), aged 3 to 16 years, compared to a control group matched to gender, age and ethnic group. Both groups were examined as to the caries index, amount of dental plaque, gingivitis, enamel defects and microbiological evaluation. The results revealed higher frequency of caries-free children in the study group. Children with craniosynostosis presented a greater amount of dental plaque, which the authors assigned to the difficult oral

hygiene control due to tooth crowding, leading them to highlight the importance of regular professional dental prophylaxis. The study group presented higher frequency of gingivitis in permanent teeth. Both groups presented similar results as to the enamel defects and microbiological evaluation.

According to Wagaiyu and Ashley[4] (1991), gingival inflammation seems to be more severe in mouth breathers, regardless of the accumulation of plaque[37]. Previous studies have also demonstrated that tooth crowding may predispose to gingivitis, regardless of the amount of plaque[4] and only in mouth breathers, providing another explanation for the greater gingival inflammation at the anterior region, where tooth crowding is more intense. Thus, there may be a combination of factors, namely mouth breathing and tooth crowding, influencing the greater gingival inflammation at the anterior region[13].

However, most studies in the literature address the periodontal parameters of non-syndromic populations, such as the studies by Anerud, et al.[3] (1983), Ashley, et al.[4] (1998) and Baelum, et al.[5] (1986). Brown, et al.[8] (1989) evaluated the role played by periodontal disease on tooth loss and observed that only 15% of the American population did not present any sign of periodontitis. Gingivitis was found in 50% of the population, moderate periodontal pockets in 33%, and severe periodontitis in 8% of the population. The molars were the teeth most frequently affected by periodontal disease and the incisors were the least affected, especially the maxillary central incisors. The authors further analyzed the symmetry of distribution of diseases among quadrants, comparing the right and left sides of maxillary and mandibular dental arches. Bilateral symmetry was more significant in young individuals. Elderly individuals exhibited less symmetry, probably due to the higher percentage of missing teeth.

Studies on the Brazilian population reveal diverse results as to the prevalence of periodontal disease. Susin, et al.[34,35] (2004, 2005) evaluated adult individuals and observed high prevalence of moderate to severe pockets and attachment loss. Almeida, et al.[1,2] (2007, 2008) evaluated the periodontal status of young and adult individuals with congenital malformations (clefts) and observed that the attachment loss is similar compared to individuals without clefts.

The concern to investigate the periodontal status of individuals with syndromes is present in the world literature, yet the publications are scarce and usually include only few cases[7,38].

Due to the rare occurrence of these syndromes[16,17,24], few studies on the oral health status of these individuals are available in the literature. For the same reason, most publications available so far are limited to case reports[16], or include small sample sizes[13,14]. Conversely, investigations on reasonably large samples include broad, heterogeneous age ranges[21,24,27,33].

The periodontal status of individuals with syndromic craniosynostosis is even more obscure, with lack of data in the literature. Thus, this study analyzed the periodontal health status of individuals with Apert, Crouzon, Pfeiffer and Saethre-Chotzen syndromes before and after toothbrushing, in addition to the efficiency of plaque removal by comparison before and after toothbrushing.

MATERIAL AND METHODS

This study was conducted at the Hospital for Rehabilitation of Craniofacial Anomalies (HRAC/USP) of the University of São Paulo, in Bauru, Brazil, following the guidelines of the Hospital's Institutional Review Board and was conducted in accordance with the World Medical Association Declaration of Helsinki. Searching a database with over 56,000 patients registered at the Hospital, 136 patients were found with a diagnosis of craniosynostosis syndromes (Table 1).

The sample was composed of 27 individuals of both genders aged 11 to 36 years, being 9 with Crouzon syndrome,

Craniosynostosis syndrome	Oral characteristics
Apert syndrome	maxillary hypoplasia, lateral palatal swellings, gingival hypertrophy, multiple tooth agenesis, shovel-shaped incisors, high caries prevalence, early tooth loss, difficult oral hygiene control due to hand malformations[13, 14, 20, 21, 24.]
Crouzon syndrome	maxillary hypoplasia, lateral palatal swellings, reduced maxillary length, maxillary hypoplasia, counterclockwise mandibular rotation, mandibular prognathism due to positional changes with normal mandibular growth, ectopic eruption, tongue thrusting, partial tooth agenesis[9, 10,11, 29, 36].
Pfeiffer syndrome	maxillary hypoplasia, lateral palatal swellings, mandibular prognathism, high-arched palate, tooth crowding[13].
Saethre-Chotzen syndrome	maxillary hypoplasia, lateral palatal swellings, narrow palate, cleft palate, Class III malocclusion, teeth with large crowns and thin and long roots, multiple pulp stones[16].

FIGURE 1- Oral characteristics of craniosynostosis syndromes

TABLE 1- Entire sample of patients with craniosynostosis syndromes registered at HRAC-USP up to years 2005 and 2006

Craniosynostosis syndromes	Death	Age <11 years	Treatment	Sample available for the study
Apert syndrome	10	37	18	9
Crouzon syndrome	0	14	10	9
Pfeiffer syndrome	1	4	3	4
Saethre-Chotzen syndrome	0	8	4	5
Total	11	63	35	27

9 with Apert syndrome, 4 with Pfeiffer syndrome and 5 with Saethre-Chotzen syndrome. The inclusion criteria comprised patients registered and treated at HRAC/USP. Individuals previously submitted to orthodontic or orthognathic treatment, serial tooth extraction or presenting with delayed neuropsychomotor development were excluded.

Clinical examinations were performed by a single examiner. Ten individuals treated at the hospital were examined twice at a one-week interval, revealing an intra-examiner agreement higher than 89% (kappa test).

The study was conducted in a two-year period. Each individual was examined only once, without further sessions for comparative evaluation. Individuals with some syndromes, such as the Apert syndrome, attend the hospital every 6 months, while individuals with other syndromes attend the hospital less frequently.

All periodontal parameters were evaluated using a periodontal probe model COLOR CODED PROBE CP-ISUNC-PCPUNC 15 (Hu-Friedy, USA), as follows:

a) Probing depth at 6 sites in each tooth (mesial, center and distal of the buccal and lingual aspects) with aid of periodontal probes. The gingival margin was taken as reference point for reading of values during probing[30].

b) Periodontal clinical attachment level evaluated at 6 sites in each tooth (mesial, center and distal of the buccal and lingual aspects) with aid of periodontal probes. The cementoenamel junction was taken as reference point for reading of values during probing[30].

c) Gingival index according to Löe[25] (1967).

d) Plaque index, described by Silness and Löe[32] (1964), which comprises visual clinical evaluation of each tooth on the mesial, distal, buccal and lingual aspects.

e) Amount of keratinized mucosa. The reference point was taken at the central region of the buccal aspect of all teeth, considering a keratinized mucosa width of = 2mm as adequate[22].

f) O'Leary plaque index according to O'Leary[28] was evaluated before and after non-supervised manual toothbrushing, on the same day, by assessment of the presence of soft debris on tooth surfaces and dento-gingival junction as well as toothbrushing efficacy. Plaque disclosure was performed with a chewable tablet (Replasul "C", Iodontosul, Porto Alegre, Brazil). This index was evaluated twice, before and after toothbrushing as routinely performed by the patients, in order to verify if the technique employed by them was effective for plaque removal. If the plaque index remained high after toothbrushing, the patient received oral hygiene instructions by the examiner.

The periodontal parameters were evaluated before toothbrushing. The plaque index was assessed before and immediately after toothbrushing, on the same day. The patients performed toothbrushing as they routinely do, followed by oral hygiene instructions and supervised toothbrushing when required.

Comparison among syndromes as to the probing depth, attachment level, gingival index, plaque index of Silness and Löe[32] and keratinized mucosa was performed by three-way analysis of variance with repeated measurements[39], for the following three factors: *syndrome* (Apert, Crouzon, Pfeiffer and Saethre-Chotzen), *dental arch* (maxillary and mandibular) and *region* (maxillary and mandibular posterior right, maxillary and mandibular anterior, and maxillary and mandibular posterior left). Comparison of the O'Leary plaque index[28] was performed by the same model, yet with addition of the factor *period* (before and after toothbrushing), at a significance level of 5%.

RESULTS

Probing depth

Table 2 reveals that only the factors *dental arch* and *region* were significant (p=0.004 and p<0.001, respectively). Application of the multiple-comparison test for each factor demonstrated that the mandibular dental arch presented significantly lower probing depth than the maxillary dental arch, and that the anterior region presented significantly lower probing depth than the right and left regions (p<0.001 in both).

Attachment level

Table 3 demonstrates that only the factor *region* was statistically significant (p <0.001). The mean attachment level was significantly lower at the anterior region compared to the right and left sides (p=0.001 and p=0.007, respectively).

Gingival index

The severity of gingival inflammation was evaluated by the gingival index. Table 4 shows an interaction effect between the factors *dental arch* and *syndrome* (p=0.037), thus requiring two types of comparison. The first presents

TABLE 2- Comparison of probing depth according to region and syndrome (ANOVA)

Factors	SS	df	MSE	F	p
Region	3.80	2	1.90	30.23	0.000*
Region*Syndrome	0.14	6	0.02	0.36	0.901
Error (Region)	2.89	46	0.06		
Syndrome	6.78	3	2.26	2.62	0.075
Error	19.81	23	0.86		

Region		Mean difference	Standard error	p	Confidence interval (95%)	
A	B	A-B			Lower limit	Upper limit
Right	Anterior	0.36	0.05	0.000*	0.24	0.47
Right	Left	0.02	0.05	1.000	-0.12	0.16
Anterior	Left	-0.33	0.05	0.000*	-0.47	-0.19

*Statistically significant at p<0.05; SS: sum of squares; df: degrees of freedom; MSE: mean square error.

TABLE 3- Comparison of attachment level according to region and syndrome (ANOVA)

Factors	SS	df	MSE	F	p
Region	2.73	2	1.37	11.31	0.000*
Region*Syndrome	0.59	6	0.10	0.81	0.570
Error (Region)	5.56	46	0.12		
Syndrome	2.93	3	0.98	0.72	0.553
Error	31.36	23	1.36		

Region		Mean difference	Standard error	p	Confidence interval (95%)	
A	B	A-B			Lower limit	Upper limit
Right	Anterior	0.33	0.08	0.001*	0.13	0.53
Right	Left	0.10	0.07	0.489	-0.08	0.28
Anterior	Left	-0.23	0.07	0.007*	-0.41	-0.06

*Statistically significant at p<0.05; SS: sum of squares; df: degrees of freedom; MSE: mean square error.

TABLE 4- Comparison of gingival index according to region and syndrome

Factors	SS	df	MSE	F	p
Region	0.15	2	0.08	1.53	0.228
Region*Syndrome	0.18	6	0.03	0.60	0.727
Error (Region)	2.31	46	0.05		
Syndrome	0.81	3	0.27	0.51	0.680
Error	12.24	23	0.53		

*Statistically significant at p<0.05; SS: sum of squares; df: degrees of freedom; MSE: mean square error.

the comparison of the types of syndromes for each region, and the second displays the comparison of the dental arches for each syndrome. The result revealed that there was no statistically significant difference between the syndromes, yet there was difference between the dental arches for the group with Apert syndrome: the mandibular dental arch

presented lower mean gingival index than the maxillary dental arch (p=0.003) and mean difference of 0.25 with confidence interval between 0.10 and 0.41.

Plaque index of Silness and Löe

Only the factor *region* was statistically significant (p=0.001) for the variable plaque index. Application of multiple-comparison test revealed statistically significant difference between the left side and the other regions. In both cases, the left region presented higher plaque index than the right and anterior regions (p=0.001 and p=0.005, respectively) (Table 5).

Keratinized mucosa

Analysis of variance demonstrated that only the factor *dental arch* was statistically significant (p=0.006). Table 6 shows that the mean amount of keratinized mucosa was statistically greater in the maxillary dental arch (p=0.006).

O'Leary plaque index

Analysis of variance (Table 7) revealed the presence of statistically significant interaction between the factors *period, dental arch* and *syndrome*, as well as between the factors *dental arch* and *region*. Therefore, we preferred to perform *post-hoc* comparisons between the syndromes by fixating the other values (Table 6 and Table 7) and comparisons between periods by fixating the other values (Table 7). These comparisons allowed the following statements:

· Before toothbrushing, in the mandibular dental arch,

TABLE 5- Comparison of plaque index according to region and syndrome

Factors	SS	df	MSE	F	p
Region	1.42	2	0.71	8.72	0.001*
Region*Syndrome	0.92	6	0.15	1.89	0.104
Error (Region)	3.76	46	0.08		
Syndrome	3.82	3	1.27	1.01	0.407
Error	29.01	23	1.26		

Region		Mean difference	Standard error	p	Confidence interval (95%)	
A	B	A-B			Lower limit	Upper limit
Right	Anterior	0.13	0.07	0.214	-0.05	0.31
Right	Left	-0.11	0.03	0.001*	-0.18	-0.04
Anterior	Left	-0.24	0.07	0.005*	-0.42	-0.07

*Statistically significant at p<0.05; SS: sum of squares; df: degrees of freedom; MSE: mean square error.

TABLE 6- Comparison of keratinized mucosa according to region and syndrome

Factors	SS	df	MSE	F	p
Region	4.49	2	2.24	2.04	0.141
Region*Syndrome	6.36	6	1.06	0.97	0.459
Error (Region)	50.56	46	1.10		
Syndrome	13.61	3	4.54	0.59	0.627
Error	176.51	23	7.67		

Region		Mean difference	Standard error	p	Confidence interval (95%)	
A	B	A-B			Lower limit	Upper limit
Right	Anterior	0.13	0.07	0.214	-0.05	0.31
Right	Left	-0.11	0.03	0.001*	-0.18	-0.04
Anterior	Left	-0.24	0.07	0.005*	-0.42	-0.07

*Statistically significant at p<0.05; SS: sum of squares; df: degrees of freedom; MSE: mean square error.

TABLE 7- Comparison of the O'Leary plaque index according to period, region and syndrome

Factors	SS	df	MSE	F	p
Period	13.53	1	13.53	98.00	0.000*
Period*Syndrome	2.05	3	0.68	4.95	0.008*
Error (Period)	3.18	23	0.14		
Period*Region	0.01	2	0.01	0.21	0.815
Period*Region*Syndrome	0.12	6	0.02	0.82	0.563
Error (Period*Region)	1.16	46	0.03		
Syndrome	4.91	3	1.64	4.10	0.018*
Error	9.18	23	0.40		

*Statistically significant at p<0.05; SS: sum of squares; df: degrees of freedom; MSE: mean square error.

TABLE 8- Comparison of the O'Leary plaque index according to syndrome, dental arch and region

Syndrome	Dental arch	Region	Mean difference (before/after toothbrushing)	Standard error	p	Confidence interval (95%)	
						Lower limit	Upper limit
Apert	Md	Right	0.48	0.11	0.000*	0.26	0.70
		Anterior	0.42	0.10	0.000*	0.22	0.62
		Left	0.33	0.09	0.002*	0.14	0.52
	Mx	Right	0.54	0.09	0.000*	0.35	0.74
		Anterior	0.46	0.08	0.000*	0.29	0.63
		Left	0.45	0.10	0.000*	0.23	0.66
Crouzon	Md	Right	0.60	0.11	0.000*	0.38	0.82
		Anterior	0.63	0.10	0.000*	0.43	0.83
		Left	0.57	0.09	0.000*	0.38	0.76
	Mx	Right	0.48	0.09	0.000*	0.28	0.67
		Anterior	0.51	0.08	0.000*	0.34	0.68
		Left	0.41	0.10	0.001*	0.19	0.62
Pfeiffer	Md	Right	0.03	0.16	0.847	-0.30	0.36
		Anterior	0.17	0.14	0.260	-0.13	0.47
		Left	0.05	0.14	0.711	-0.24	0.34
	Mx	Right	0.09	0.14	0.512	-0.20	0.39
		Anterior	0.14	0.12	0.270	-0.11	0.39
		Left	0.23	0.16	0.145	-0.09	0.56
Saethre-Chotzen	Md	Right	0.67	0.14	0.000*	0.37	0.96
		Anterior	0.81	0.13	0.000*	0.54	1.08
		Left	0.75	0.12	0.000*	0.50	1.01
	Mx	Right	0.52	0.13	0.000*	0.26	0.78
		Anterior	0.49	0.11	0.000*	0.26	0.72
		Left	0.63	0.14	0.000*	0.35	0.92

*Statistically significant at p<0.05; Md: mandibular; Mx: maxillary.

in the right and anterior regions, the Pfeiffer syndrome presented significantly lower mean than all other syndromes. In the left side, the Pfeiffer syndrome presented significantly lower mean than all other syndromes except for Apert syndrome (p=0.092, Table 8);

· Before toothbrushing, in the maxillary dental arch, the group with Pfeiffer syndrome demonstrated significantly lower mean than all other groups only at the anterior region (Table 8). In the right side, there was difference only between the Pfeiffer and Crouzon syndromes, with mean 0.62 units lower for the Pfeiffer group compared to the Crouzon group (p=0.017);

· After toothbrushing, there was no statistically significant difference between the syndromes;

· Comparison of periods, fixating the other factors, revealed statistically significant differences for all syndromes, except for Pfeiffer syndrome (Table 8).

DISCUSSION

Knowledge on the inherent oral changes of individuals with syndromic craniosynostosis is very important for oral health maintenance of these individuals because some of their structural characteristics may influence the homeostasis of oral tissues.

Analysis of results according to the region aimed to verify if some oral characteristics of these individuals, such as tooth crowding, maxillary hypoplasia, and others, might influence the periodontal health. The mean probing depths and clinical attachment levels were higher for the posterior sextants for all syndromes. This greater severity in posterior teeth is also observed in non-syndromic individuals[15,18].

The difficult access to the posterior teeth may have been worsened by the presence of gingival hyperplasia, inherent to these individuals, especially on the lingual aspects[17,24,33].

Gingival inflammation was uniformly distributed among regions and syndromes. Some factors, such as gingival hyperplasia and mouth breathing, may influence as well. Mouth breathers may present greater inflammation at the anterior region, regardless of plaque accumulation[19,37]. Tooth crowding should also be considered as a factor that may predispose to gingivitis[4].

Analysis of the plaque index revealed statistically significant difference between the left side and the other regions. This might be explained by the fact that most individuals are right-handed, which would impair the access to and visibility of the opposite side. In both cases, the left region exhibited higher plaque index than the right and anterior regions. Even though dental plaque is the cause of periodontal disease, populations of developing countries do not necessarily exhibit higher prevalence and severity of periodontal disease, despite the greater presence of plaque and gingivitis[5,6,23,26,31]. The progression of gingivitis to periodontitis does not occur in a linear manner. In some cases, the lesions are restricted to the gingival tissues, without loss of connective attachment or alveolar bone. In other cases, they progress rapidly to destructive periodontitis, with loss of supporting periodontal tissue.

Gingival hyperplasia, especially at the palatal region, is inherent to some syndromes. This study evaluated the keratinized mucosa width on the buccal aspect to check if such change also affected this region. There were no significant differences between the keratinized mucosa width and region or syndrome, which indicates that gingival hyperplasia probably does not affect the buccal aspect.

Considering that the results observed by assessment of the O'Leary plaque index were significantly lower for patients with Pfeiffer syndrome, these individuals may present milder syndromic characteristics than the other individuals, with greater manual dexterity and minor oral changes, such as lateral palatal swellings and tooth crowding.

CONCLUSIONS

Within the limitations of the present study due to the small sample size, the periodontal status of individuals with Apert, Crouzon, Pfeiffer and Saethre-Chotzen syndrome seems to be similar for most variables analyzed. The posterior region was the most affected by tissue changes for all syndromes. Individuals with Pfeiffer syndrome presented greater toothbrushing efficiency those with the other syndromes evaluated in this study.

Further studies on larger and randomized samples should be conducted. These patients should be followed longitudinally to check the evolution or not of periodontal disease.

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