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Original Article

Surgical effects of rehabilitation protocols on dental arch occlusion of children with cleft lip and palate

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Aim: to evaluate the surgical effects of two rehabilitation protocols on dental arch occlusion of 5-year-old children with or without cleft lip and palate. **Methods:** this is a retrospective longitudinal study the sample comprised 45 digitized dental casts divided into followed groups: Group 1 (G1) – children who underwent to cheiloplasty (Millard technique) at 3 months and to one-stage palatoplasty (von Langenbeck technique) at 12 months; Group 2 (G2) – children who underwent to cheiloplasty (Millard technique) and two-stage palatoplasty (Hans Pichler technique for hard palate closure) at 3 months and at 12 months to soft palate closure (Sommerlad technique); and Group 3 (G3) – children without craniofacial anomalies. Linear measurements, area, and occlusion were evaluated by stereophotogrammetry software. Shapiro-Wilk test was used to verify normality. ANOVA *followed by posthoc* Tukey test and Kruskal-Wallis *followed by posthoc* Dunn tests were used to compared groups. **Results:** For the measures intercanine distance (C-C'), anterior length of dental arch (I-CC'), and total length of the dental arch (I–MM'), there were statistical differences between G1x G3 and G2xG3, the mean was smaller for G1 and G2. No statistically significant differences occurred in the intermolar distance and in the dental arch area among groups. The occlusion analysis revealed significant difference in the comparison of the three groups (p=0.0004). **Conclusion:** The surgical effects of two rehabilitation protocols affected the occlusion and the development of the anterior region of the maxilla of children with oral clefts when compared to children without oral clefts.

Keywords: Cleft lip. Cleft palate. Dental arch. Imaging, three-dimensional. Dental occlusion.

Introduction

The individual with cleft lip and palate undergoes a complex rehabilitative treatment through primary plastic surgeries, namely cheiloplasty and palatoplasty[1]. These surgical procedures aim to rehabilitate and return the proper speech, hearing, and masticatory functions, directly influencing the self-esteem and social-affective integration of individuals with oral clefts[2]. Different rehabilitation protocols have been used over the years to repair the lip and palate[3].

To understanding the outcomes of plastic surgery and searching for suitable technical approach to decrease the iatrogenic effects of the rehabilitative procedures are essential to the rehabilitation of individuals with oral clefts, and they provide more favorable results that would consequently improve the quality of life[4]. The main aspects of the clef lip and palate repair is to understand the outcomes of different rehabilitative procedures. The rehabilitation starts with the closure of the lip. One of the most techniques used is Millard's, which consists in incisions that allow the rotation of the flap for lip closure[5]. Von Langenbeck's technique, a procedure for close palate, requires relaxing incisions to promote union of the muscles at the level of the septum[6,7]. Another technique for hard palate repair that can be used is Hans Pichler, which consist in the closure using a vomer flap[8]. Sommerlad's technique, is a procedure to enhance velopharyngeal competence, the performance is to reposition and reinserted of the elevated muscle of the soft palate, in posterior edge of hard palate, in order to reestablish the muscle complex, contributed to the function of the soft palate[9].

The differences in protocols can improve maxillary growth in cleft patients, and its importance is related to an achievement of the best rehabilitative protocol, since there is no gold standard protocol for cleft patient. The protocols are performed by the experience of the surgeon or for the convenience of the type, extension of the cleft. This justifies the evaluation and comparison of the dental arch development and the impact of the different surgical protocols in 5-year-old children. This study null hypothesis is that the dental arch morphology of children undergoing different rehabilitation surgical protocols is not statistically different from that of children without oral clefts, thus, the dental arch with patients with cleft remain without any restriction after primary surgeries, being as the same pattern with match-control-peers. Thus, this study aimed to evaluate the surgical outcomes of two rehabilitation protocols on dental arch occlusion of 5-year-old children with or without cleft lip and palate.

Materials and Methods

The approved protocol by the Institutional Review Board is CAAE: 40034620.6.0000.5441. This is a retrospective longitudinal study, in the period of 2010 to 2019. The study the sample comprised 45 digitized dental casts divided into the three different groups: Group 1 (G1) – children submitted to cheiloplasty (Millard technique) at 3 months and one-stage palatoplasty (von Langenbeck technique) at 12 months; Group 2 (G2) – children submitted to cheiloplasty (Millard technique) and two-stage palatoplasty (Hans Pichler technique for hard palate closure) at 3 months and 12 months to soft palate closure (Sommerlad technique); and Group 3 (G3) – children without craniofacial anomalies (control group).

Sample size estimative was accomplished according to the study of Maulina et al.[10] (2007). We considered a standard deviation of 2.73 millimeter (mm) in the intercanine distance of children with unilateral cleft lip and palate, the level of significance used was 5%, where p≤0.05 was considered significant, power test of 80%, and the clinically minimum difference to be detected of 2.95 mm. The minimum sample size for each group was 14 children.

Inclusion criteria (G1 and G2) comprised maxillary dental casts of children from 5 years old, with unilateral cleft lip and palate, and without other craniofacial anomalies, with complete primary dentition, operated by the same plastic surgeon during their first year of life, at the rehabilitative center. The exclusion criteria (G1 and G2) were syndrome or other associated malformations, uncooperative children, and absence of the maxillary primary canines and/or second molars. Inclusion criteria (G3) comprised maxillary dental casts of children from 5 years old, without cleft lip and palate and with complete primary dentition, at a Dental School University. The exclusion criteria (G3) were absence of the maxillary primary canines and/or second molars.

The analyzed images was obtained from digitized dental casts by a three dimensional (3D)D scanner (Scanner R700™ Scanner; 3Shape AS, Copenhagen, Denmark), and the digitized images were analyzed by two examiners in the stereophotogrammetry software (Mirror imaging software, Canfield Scientific, Inc., Fairfield, NJ, USA)[11–13].

The linear measurements were evaluated: intercanine distance (C–C') – transversal line between the cusps of the maxillary left and right primary canine; intermolar distance (M–M') – transversal line between the distal points of the palatal surface of the primary second molars; anterior dental arch length (I–CC') – straight line passing from the interincisive point (I) perpendicularly to the C–C' distance; and total dental arch length (I–MM') – straight line from the point (I) perpendicularly to the distance M–M'[14]. The linear measurements were quantified in mm, Figure 1. The palate area

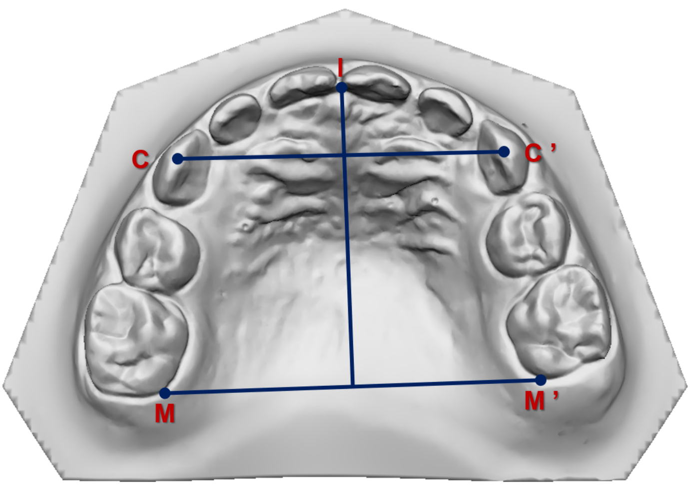

Figure 1. Linear measurements.

was marked by points passing through the palatal surfaces of the teeth. The posterior limit of the dental arch was the distance M–M'[10], Figure 2. The area was quantified in square millimeters (mm^2).

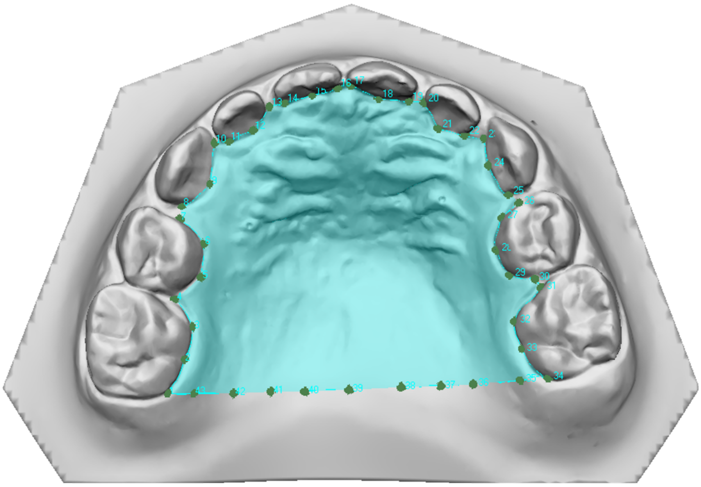

Figure 2. Palate area.

The three-dimensional images of the models in occlusion were evaluated by the index of Atack et al.[15] (1997). This index defines the systematization criteria for quantifying the occlusion morphology in individuals with unilateral cleft lip and palate, ranging from 1 to 5. The greater the index, the greater is the severity of the occlusion considering the interarch relationship, the maxillary arch shape, and the tipping of the maxillary incisors (Table 1).

Table 1. Classification of the index of Atack.

Index	Description	Prognosis
1	Positive overjet. Normal or palatal tipping of the maxillary incisors. Lack of open or crossbite.	Excellent
2	Positive overjet. Normal or labial tipping of the maxillary incisors. Tendency towards crossbite and unilateral crossbite. Tendency towards open bite at the cleft side.	Good
3	Anterior edge-to-edge bite. Labial tipping of the maxillary incisors or overjet with palatal tipping of the incisors. Tendency towards open bite at the cleft side.	Regular
4	Negative overjet. Normal or labial tipping of the maxillary incisors. Tendency towards open bite at the cleft side. Tendency towards posterior unilateral or bilateral crossbite.	Poor
5	Negative overjet. Labial tipping of the maxillary incisors. Bilateral crossbite.	Very poor

All the statistical analyses were performed by GraphPad Prism software (Prism 5 for Windows - Version 5.0 – GraphPad software., Inc. San Diego, USA), with the level of significance used was 5%, where $p \leq 0.05$ was considered significant. The normality of the samples were analyzed by Shapiro-Wilk test. To check the method reliability, 1/3 of the sample was evaluated twice with a 15-day interval[12,13]. Wilcoxon test verified the intraexaminer analysis, while Mann-Whitney test verified the interexaminer analysis. Dahlberg's formula quantified the causal error. ANOVA and *posthoc* Tukey test, Kruskal-Wallis and *posthoc* Dunn tests were used to compared all three groups.

Results

The sample was comprised by 16 children in G1, 14 children in G2, and 15 children in G3, totalizing 45 evaluated dental casts. The study participants mean age was 6.08 (± 0.65) years (Table 2). Both the linear measurements and the area revealed no statistically significance in intraexaminer (Wilcoxon test, p= 0.114 and Dahlberg's formula = 0.829) and interexaminer analyses (Mann-Whitney test, p=0.579). The occlusion analysis revealed no statistically significance differences in intraexaminer (Wilcoxon test, p = 0.423) and interexaminer analyses (Mann-Whitney test, p=0.983).

Table 2. Statistical analysis of sample.

Parameters	Group 1	Group 2	Group 3	P-value (test)
Male / Female (n)	9 / 7	11 / 3	10 / 5	0.433 (Chi-square)
Age (Years)	6.93	5.92	5.39	0.367 (Kruskal-Wallis)

For the measures intercanine distance (C-C'), anterior length of dental arch (I-CC'), and total length of the dental arch (I–MM'), there were statistical differences between G1x G3 and G2xG3, the mean was smaller for G1 and G2. There was no significant differences presented in the intermolar distance (M–M') and in the dental arch area among groups (Table 3).

Table 3. Intergroup analysis of the anthropometry of the dental arches (ANOVA post-hoc Tukey test).

Analyses	Unit	Group 1 Mean	SD	Group 2 Mean	SD	Group 3 Mean	SD	*P*
C–C'	mm	25.29 A	3.90	25.04 A	3.01	29.90 B	1.56	**<0.0001***
I–CC'	mm	5.19 A	2.51	5.78 A	1.27	6.87 B	0.86	**0.034***
M–M'	mm	35.31 A	3.99	35.06 A	2.39	36.29 A	1.59	0.425
I–MM'	mm	24.04 A	3.84	23.96 A	1.36	26.95 B	1.29	**0.002***
Area	mm²	788.23 A	144.65	826.27 A	77.64	845.28 A	98.56	0.361

* Statistically significant difference. SD: Standard deviation.
Different capital letters in line means statistically significant difference.

Table 4 shows the occlusion analysis of G1, G2, and G3 according to the index of Atack. The intergroup comparison showed no statistically significant differences for G1 *vs.* G2, but statistically significant differences for G3 *vs.* G1 and G3 *vs.* G2.

Table 4. Classification of the index of Atack by group. Intergroup analysis of the index of Atack (Kruskal-Wallis test *post-hoc* Dunn test).

Index	Group 1 n (%)	Group 2 n (%)	Group 3 n (%)	*P*
1	1 (6.25)	0 (0)	7 (46.67)	
2	6 (37.50)	4 (28.75)	6 (40)	
3	0 (0)	5 (35.71)	2 (13.33)	
4	6 (37.50)	4 (28.57)	0 (0)	
5	3 (18.75)	1 (7.14)	0 (0)	
Total	16 (100) A	14 (100) A	15 (100) B	**0.0004***

* Statistically significant difference.
Different capital letters in line means statistically significant difference.

Discussion

This present study justifies in the attempted to understand better the differences between two different rehabilitative protocol, highlighting all the children were operated by one surgeon. In this way, the outcomes can be more favorable for the comprehension of what is important in the rehabilitative process.

This present study exhibited a greater measurement of the intercanine distance, anterior dental arch length, and total dental arch length for children without clefts. The rationale behind this finding would be the restriction initiated by primary surgeries in the anterior (canine area) and anterior-posterior transversal growth. There were no significant differences in the intermolar distance and area. Thus, it can be affirmed that the primary surgeries did not change the posterior transversal growth and the dental arch area. By corroborating with the maxillary restriction caused by the primary surgeries, Bruggink et al.[16](2019), evaluated longitudinally individuals without oral clefts and followed the maxillary growth through the first year of life and remarkably found that the rate growth between the canines increased between 3 and 6 months of life. Previous study analyzed the maxillary dimensions at the first six months of life and estimated that the relative transversal growth of the anterior portion of the maxilla is around three times quicker than that of the posterior portion (81.9% *vs.* 26.2%)[17]. This points out to an anterior widening of the maxilla during that period and highlights the impact of cheiloplasty performed at the first months of life. Moreover, the treatment prognosis is categorized by the cleft amplitude severity, highlighting the width size. The bigger the cleft size, the greater is the probability of the healing tissue negatively impact on the maxillary growth[18]. The study of Huang et al.[19](2002), evaluated the maxilla of individuals with unilateral cleft lip and palate and found an increasing in the dental arch linear measurements after a period of 12 months, except for the anterior region that displaced towards palatine after the cheiloplasty.

The method to obtain these measurements is very important. According to Kongprasert et al.[20](2019), three-dimensional evaluation has better accuracy and validity than two-dimensional evaluation, and it performances an important role in the follow-up of the change in dental arch dimensions towards all directions. Digitized models have the advantages of construction and analysis, absence of damage, that is, preservation of the dental casts. Thus, digitized dental cast has replaced dental casts as gold-standard[20]. Previous studies reported the validity of the 3D stereophotogrammetry, including the clinical environment[21].

The occlusal analysis by the Index of Atack is performed at 5 years-old because this is the age children are at complete deciduous denture. This index is measured in a scale ranging from 1 to 5, seeing that the greater the index, the worst is the facial profile, oscillating from regular occlusion to anterior and/or posterior crossbite[15]. The study of dental casts plays a relevant therapeutic role in the treatment of individuals with oral clefts because it points out the dimensional alterations and enables the use of indexes regarding treatment[22].

This present study showed no statistically differences between the groups with clefts. This may suggest the interference of the different primary surgeries techniques on the occlusion development. The impact of the primary surgeries is still difficult to measure, which one is more suitable for the growth[23]. Indeed, the literature lacks comparative studies on the occlusal analysis in children with and without cleft lip and palate. Thus, the comparison with children without clefts revealed that different surgical techniques directly influenced on the occlusal outcome. This result may contribute with the elaboration of a satisfactory rehabilitation protocol.

The potential strength of this study is the sample, because all the patient present in this study was operated by the same surgeon, so the sample have no operator bias, however this limits the sample number. A limitation issue that can be pointed is the size of the cleft before primary surgeries, this can be an important challenge for the surgeon, because wider is the cleft, more soft tissue is needed and more in the potential of scar and retraction. Therefore, further studies can be delineated in relation of cleft width.

In conclusion, the surgical effects of two rehabilitation protocols affected the occlusion and the development of the anterior region of the maxilla of children with oral clefts when compared to children without oral clefts.

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Data availability

Datasets related to this article will be available upon request to the corresponding author.

Conflict of Interests

None.

Author contribution

Paula Karine Jorge – have made substantial contributions to conception and design, or acquisition of data, or analysis and interpretation of data. And have been involved in drafting the manuscript or revising it critically for important intellectual content

Níkolas Val Chagas – Contributed substantially to the conception and design of the study, the acquisition of data, and the analysis and interpretation

Eloá Cristina Passucci Ambrosio - Drafted or provided critical revision of the article, the analysis and interpretation, and provided final approval of the version to publish

Cleide Felício Carvalho Carrara – have given final approval of the version to be published

Fabrício Pinelli Valarelli – have been involved in drafting the manuscript or revising it critically for important intellectual content

Maria Aparecida Andrade Moreira Machado - agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Thais Marchini Oliveira - Drafted or provided critical revision of the article. Provided final approval of the version to publish. Agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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