UNIVERSIDADE DE SÃO PAULO
HOSPITAL DE REABILITAÇÃO DE ANOMALIAS CRANIOFACIAIS

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Evaluation of palatal volume in children with cleft lip and palate: a comparison of two surgical protocols

Avaliação do Volume dos arcos dentários de crianças com fissura labiopalatina operadas por dois protocolos cirúrgicos

BAURU

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Dissertação constituída por artigo apresentada ao Hospital de Reabilitação de Anomalias Craniofaciais da Universidade de São para obtenção do título de Mestre em Ciências da Reabilitação, na área de concentração Fissuras Orofaciais e Anomalias Relacionadas.

Orientadora: Profa. Dra. Thais Marchini de Oliveira.

Versão Corrigida

BAURU

2021

Ribeiro Homem Alcarde, Amanda

Evaluation of palatal volume in children with cleft lip and palate: a comparison of two surgical protocols/ Amanda Ribeiro Homem Alcarde – Bauru, 2021.
33 p. : il.

Dissertação (mestrado) -- Hospital de Reabilitação de Anomalias Craniofaciais, Universidade de São Paulo, ano de defesa.

Orientador: Prof. Dr. Thais Marchini de Oliveira

Nota: A versão original desta dissertação/tese encontra-se disponível no Serviço de Biblioteca e Documentação da Faculdade de Odontologia de Bauru - /FOB/USP.

Comitê de Ética do HRAC-USP

Protocolo nº:

ERRATA

FOLHA DE APROVAÇÃO

DEDICATÓRIA

Dedico esse projeto aos meus pais, **Reynaldo** *(in memoriam)*, e minha mãe **Angela Glaucia**, que sempre apoiaram meus sonhos e projetos e fizeram tudo para que eu tivesse os melhores estudos e pudesse realizar sonhos como esse. Esta dissertação é a prova de que todo o investimento e dedicação de vocês, valeram a pena.

Dedico este projeto de pesquisa ao meu marido **Guilherme**, que foi meu maior incentivador, me deu todo o suporte para que eu realizasse mais essa grande etapa da minha vida. Esta é mais uma, das muitas conquistas ao seu lado. Muito obrigada.

AGRADECIMENTOS

À **Deus**, minha gratidão pela saúde e proteção.

Aos meus **irmãos**, pelo apoio durante o processo.

À minha querida Orientadora **Profa. Dra. Thais Marchini de Oliveira Valarelli**, pelo apoio, pela confiança, pela oportunidade em aprender com você, pelas valiosas e incontáveis horas dedicadas ao projeto, sempre com uma presença cheia de otimismo. Muito obrigada.

Ao **Prof. Dr. Fabrício Valarelli,** pelo apoio e incentivo.

À parceira de pesquisa **Eloá Cristina Passucci Ambrósio** pela paciência e por todo suporte.

Aos **pacientes**, sem os quais essa pesquisa não seria possível

Ao **Hospital de Reabilitação de Anomalias Craniofaciais – USP**, na pessoa do senhor superintendente Prof. Dr**. Carlos Ferreira dos Santos.**

Aos meus amigos **Isabela Sartori e Nikolas Val** pelo convívio diário e amizade.

À **Ana Regina**, da pós graduação do HRAC, que sempre me deu apoio e tirou todas as minhas dúvidas. Muito obrigada, pela sua atenção e paciência.

“*O sucesso é a soma de pequenos esforços repetidos dia após dia*.” (Robert Collier)

ABSTRACT

Evaluation of palatal volume in children with cleft lip and palate: a comparison of two surgical protocols

This study aimed to compare the palatal volume in children with unilateral cleft lip and palate before and after two surgical protocols. The sample comprised 120 digitized dental models divided into: Group 1 (G1) – children submitted to cheiloplasty at three months (Millard technique) and one-step palatoplasty at 12 months (von Langenbeck technique); Group 2 (G2): children submitted to cheiloplasty (Millard technique) and hard palate closure (Hans Pichler technique) at three months and soft palate closure at 12 months (Sommerlad technique). The dental arches were evaluated at three periods. The volume was measured through stereophotogrammetry system software. Repeated measures ANOVA followed by Tukey test and Friedman test followed by Dunn test was used for intragroup comparisons. Independent t test and Mann-Whitney test evaluated intergroup comparisons. The intragroup analysis revealed that G1 had a statistically significant increase in volume at T2 followed by a reduction at T3 (p=0.003); G2 showed a statistically significant increase of dental arch volume between T1 and T2 (p=0.001). The surgical protocol influenced the palatal volume of children with unilateral cleft lip and palate. Over time, in the same group, this study suggested that two-step palatoplasty was more suitable for dental archdevelopment.

Keywords: Cleft lip. Cleft palate. Dental arch. Imaging, Three–dimensional. Anthropometry.

RESUMO

Avaliação do volume dos arcos dentários de crianças com fissura labiopalatina operadas por dois protocolos cirúrgicos

O objetivo do presente estudo foi avaliar o volume dos arcos dentários de crianças com fissura unilateral de lábio e palato antes e após dois protocolos cirúrgicos. A amostra foi composta por 120 modelos dentários digitalizados divididos em:Grupo 1 (G1) – crianças operadas para reparo labial (técnica de Millard) aos 3 meses e palatoplastia em única etapa (técnica de von Langenbeck) aos 12 meses. Grupo 2 (G2): criançasoperadas para reparo labial (técnica de Millard) e do palato duro (técnica de Hans Pichler)aos 3 meses de vida; o reparo do palato mole (técnica de Sommerlad) foi efetuado aos 12 meses. A amostra foi avaliada em três tempos, Tempo 1 (T1) – pré cirurgias plásticas primárias; Tempo 2 (T2) – 1ª etapa pós cirurgias plásticas primárias; Tempo 3 (T3) – 2ª etapa pós cirurgias plásticas primárias. O volume foi analisado por meio do software do sistema de estereofotogrametria. A Análise de Variância (ANOVA) de medidas repetidas seguida pelo Teste de Tukey e o Teste de Friedman seguido pelo Teste de Dunn foram aplicados nas análises intragrupos; enquanto os Testes T independente e Mann-Whitney, nas comparações intergrupos (nível de significância de 5%). Na análise intragrupo, G1 apresentou diferença estatisticamente significativa com aumento do volume em T2 seguido de uma redução em T3 (p=0.003). Em G2 houve diferença estatisticamente significativa com aumento volumétrico do arco dentário entre T1xT2 (p=0.001). Conclui-se que, o volume dos arcos dentários de crianças com fissura unilateral de lábio e palato foi influenciado pelos protocolos cirúrgicos. Ao longo do tempo, no mesmo grupo, este estudo sugere que reparo do palato em duas etapas é mais adequado para desenvolvimento dos arcos dentários.

Palavras–chave: Fenda Labial. Fissura Palatina. Arco Dental. Imageamento Tridimensional. Antropometria.

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SUMMARY

1 INTRODUCTION

Oral clefts are classified by morphology (cleft lip, cleft lip and palate, and clef palate) position (unilateral, bilateral, and median), and extension (complete and incomplete) (Silva Filho et al., 1992). Individuals with oral clefts requires a multidisciplinary team to improve the functional, social, and psychological aspects of their lives because they undergone a long treatment involving different rehabilitation protocols (SHI E LOSEE, 2015; MOSMULLER *et al.*, 2013; PAPAMANOU *et al.*, 2012). The rehabilitation process is challenging, complex, and lasts since birth to adulthood. Generally, rehabilitation begins with surgical procedures to repair the functional and anatomic alterations (FREITAS *et al.*, 2012). However, these surgeries cause negative effects on the maxillary growth and midface (NAQVI *et al.*, 2015).

The rehabilitative protocol begins with the primary surgeries (cheiloplasty and palatoplasty), which aimed at restoring the face symmetry (NADJMI *et al.*, 2016) and correct the anatomic defect, improving esthetics and function to enable favorable conditions and quality of life (CARRARA *et al.*, 2016; JOKLOVÁ *et al.,* 2020). The surgeries play a paradoxical role in rehabilitation because the repair of the anatomic functional defect causes sagittal and transversal alterations in maxillofacial development. Consequently, the post-surgical healing tissue tension negatively influences on the skeletal growth of the maxilla due to a mucoperiosteal displacement that results in a thicker fibrous tissue (JONES *et al.,* 2016; RUSSELL, L. M., 2015).

Over the years, the palatal skeletal structures have been analyzed by two-dimensional (2D) methods, comprising periapical, occlusal, and panoramic radiographs (FEICHTINGER *et al.*, 2009). 2D methods have limitations, including the number of ionizing radiations, especially when longitudinal examinations are necessary in growing individuals (PUCCARELLI *et al.,* 2015). In the two last decades, many non-invasive three-dimensional (3D) studies have provided a more objective measure of the surgeryoutcome and face appearance (WONG *et al.,* 2019; AMBROSIO *et al.*, 2020), in operatedindividuals with oral clefts. These images combined with new evaluation methods increased the amount of data available for analysis (WONG *et al.,* 2019; MELLO *et al.*, 2019). The option for non-ionizing examinations includes intraoral 3D surface scanning or scanning of dental casts. Both have the advantage of not using radiation in comparisonto computed tomography (MONGA *et al.*, 2020).

Both surgeons and orthodontists adopt face stereophotogrammetry for diagnosis, planning, and treatment, as well as the evaluation of the treatment outcomes in children with oral clefts and adults submitted to orthognathic surgery (BUGAIGHISI.*et al.,* 2014;

VAN LOON, B.*et al.*, 2015).

Some researchers affirmed that the growth differences occur due to cheiloplasty and palatoplasty, while others believe these differences have embryological or genetic origin (BISHARA,1976; BARDACH,1979; ROSS, 1980; ZHENG *et al*., 2016). It is worth noting that the literature lacks information on the most suitable technique of primary surgery that would result in smaller restrictive growth effects on the dental arches of this individuals (MIKOYA *et al.*, 2015; TOME *et al.*, 2016).

The Millard technique of cheiloplasty comprises the projection of relaxing incisions that enable the flap rotation to close the lip (DEMKE; SHERARD, 2011). The most used technique for palatoplasty is von Langenbeck technique, described in 1861 (VON LANGENBECK, 1861). In this technique, relaxing incisions enable the displacement of mucoperiosteal flaps, that are sutured at nasal septum level, fixed only by the palatal vascular bundle (VON LANGENBECK, 1861; SILVA FILHO; FREITAS, 2007). In 1926, Hans Pichler included the vomer flap to close the hard palate (BOSI; BRANDÃO; YAMASHITA, 2016). Aiming at improving the velopharyngeal competency, Kriens (1969) proposed the anatomic repositioning of the palatine veil lifter muscle. This muscle is fixed at the posterior margin of the hard palate and its fibers are longitudinally directed.

Some studies have evaluated protocols with one and two-step palatoplasty and have affirmed that the total closure of the palate (one-step) before two years of age may result in earlier maxillary growth restriction (REISER; SKOOG; ANDLIN-SOBOCKI,2013). Some rehabilitation centers indicate to perform one-step palatoplasty between 12and 18 months of age to avoid growth disturbs (YONG *et al.*, 2010). Two-step palatoplastywould postpone the growth inhibition up to hard palate closure (REISER; SKOOG; ANDLIN-SOBOCKI; 2013). Notwithstanding, late palatoplasty would impair the speech development (VAN LIERDE *et al.*, 2004). Thus, the literature lacks consensus on primary surgery type, technique, and time that would result in smaller restrictive effects on maxillary growth of these individuals. (MIKOYA *et al.*, 2015; TOME *et al.*, 2016; REDDY *et al.*, 2017; FALZONI, 2019).

The analysis of the dental cast through pre-determined anatomic points markedon 3D images have been used for evaluating individuals with oral clefts (MIKOYA *et al.*, 2015; LIPPOLD *et al.*, 2015; RUSSEL *et al.*, 2015; ZHU *et al.*, 2016; CODARI *et al.*, 2016; MENEZES *et al.*, 2016; FALZONI *et al.*, 2016; SAKODA *et al.*, 2017; AMBROSIO *et al.*, 2018A; AMBROSIO *et al.*; 2018B; RANDO *et al.*, 2018; BRUGGINK *et al.*, 2019; KONGPRASERT *et al.*, 2019). The virtual evaluation of dental arch morphology is an easy procedure and improves the diagnosis and treatment planning tailored for each individual. However, the literature lacks studies comparing the dental arch volume of individuals with cleft lip and palate at the first years of life. This study aimed at gathering knowledge on the aspects interfering on the maxillary development of children with UCLP and at improving further research with new parameters and rehabilitation surgical protocols through volumetric analysis.

2 OBJECTIVE

This study aimed to compare the palatal volume in children with unilateral cleft lip and palate before and after two surgical protocols.

3 ARTICLE

3.1 ARTICLE

Evaluation of palatal volume in children with cleft lip and palate: a comparison of two surgical protocols

Abstract

This study aimed to compare the palatal volume in children with unilateral cleft lip and palate before and after two surgical protocols. The sample comprised 120 digitized dental cast divided into: Group 1 (G1) – children submitted to cheiloplasty at three months (Millard technique) and one-step palatoplasty at 12 months (von Langenbeck technique); Group 2 (G2): children submitted to cheiloplasty (Millard technique) and hard palate closure (Hans Pichler technique) at three months and soft palate closure at 12 months (Sommerlad technique). The dental arches were evaluated at three periods. The volume was measured through stereophotogrammetry system software. Repeated measures ANOVA followed by Tukey test and Friedman test followed by Dunn test was used for intragroup comparisons. Independent t test and Mann-Whitney test evaluated intergroup comparisons. The intragroup analysis revealed that G1 had a statistically significant increase in volume at T2 followed by a reduction at T3 (p=0.003); G2 showed astatistically significant increase of dental arch volume between T1 and T2 (p=0.001). The surgical protocol influenced the palatal volume of children with unilateral cleft lip and palate. Over time, in the same group, this study suggested that two-step palatoplasty was more suitable for dental arch development.

Keywords: Cleft lip. Cleft palate. Dental molds. Three-dimensional image.

Introduction

Individuals with oral clefts requires a multidisciplinary team to improve the functional, social, and psychological aspects of their lives because they undergone a long treatment involving different rehabilitation protocols [1-2]. The rehabilitation process is challenging, complex, and lasts since birth to adulthood. Generally, rehabilitation begins with surgical procedures to repair the functional and anatomic alterations [3]. However, these surgeries cause negative effects on the maxillary growth and midface [4].

The rehabilitative protocol begins with the primary surgeries (cheiloplasty and palatoplasty), which aimed at restoring the face symmetry and correct the anatomic defect, improving esthetics and function to enable favorable conditions and quality of life [5,6].

Some studies have evaluated protocols with one and two-step palatoplasty and have affirmed that the total closure of the palate (one-step) before two years of age may result in earlier maxillary growth restriction [11]. Some rehabilitation centers indicate to perform one-step palatoplasty between 12 and 18 months of age to avoid growth disturbs [12]. Two-step palatoplasty would postpone the growth inhibition up to hard palate closure[11]. Notwithstanding, late palatoplasty would impair the speech development [13]. Thus, the literature lacks consensus on primary surgery type, technique, and time that would result in smaller restrictive effects on maxillary growth of these individuals [14-17].

The analysis of the dental cast through pre-determined anatomic points markedon 3D images have been used for evaluating individuals with oral clefts [18-24]. The virtual evaluation of dental arch morphology is an easy procedure and improves the diagnosis and treatment planning tailored for each individual. However, the literature lacks studies comparing the palatal volume of individuals with cleft lip and palate at the first years of life. This study aimed at gathering knowledge on the aspects interfering on the maxillary development of children with UCLP and at improving further research with new parameters and rehabilitation surgical protocols through volumetric analysis. This study aimed to compare the palatal volume in children with unilateral cleft lip and palate before and after two surgical protocols.

Material and Methods

This study was submitted and approved by the Institutional Review Board.(CAAE: 39842920.1.0000.5441).Inclusion criteria comprised healthy children with UCLP, operated by the same surgeon, who did not begin the rehabilitative treatment. Exclusion criteria were children with other malformations and/or syndrome; absent documentation or poor-quality dental casts.

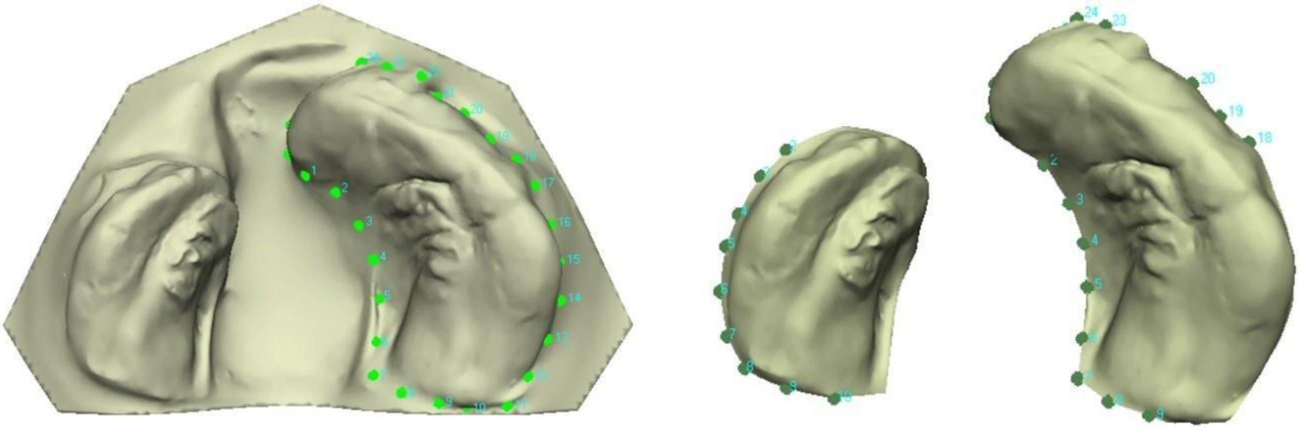
Sample size was obtained according to the study of Pucciarelli et al. (2015)[25], considering a standard deviation of 0.49 cm³ for the greater bone segment prior to the lip closure, with level of significance of 5%, test power of 80%, and the minimum difference to be clinically detected of 0.45 cm³. The minimum sample size was 20 children per group.

This present study had two groups according to the surgical protocol: Group 1 (G1) – children submitted to cheiloplasty at 3 months (Millard technique) and one-step palatoplasty at 12 months (von Langenbeck technique); Group 2 (G2): children submitted to cheiloplasty (Millard technique) and hard palate closure (Hans Pichler technique) at 3 months and soft palate closure (Sommerlad technique) at 12 months 9 (two-step palatoplasty).

The children had the impressions of the dental arch at three different periods: T1 – cheiloplasty; T2 – 1st post-surgical phase; T3 – 2nd post-surgical phase. The dental casts were digitized by a 3D scanner (Scanner R700TM Scanner; 3Shape AS, Copenhagen, Denmark). Two examiners analyzed the digitized dental arches through stereophotogrammetry software (Mirror imaging software, Canfield Scientific, Inc., Fairfield, NJ, USA) [18,20,21,25].

The analysis of the estimated volume of the dental arches was performed according to the methodology described by Pucciarelli et al. (2015)[25], and quantified in cubic centimeters (cm³). Each palatal bone segment was delimited by points through software. The points were manually marked between the alveolar edge and the maxilla (Figure 1A). The number of points were determined by the size of each segment. For each bone segment, the points were virtually projected to be separated from the dental cast base (Figure 1B).

Figure 1 – Dental arch with UCLP. **A)** Palatal bone segment delimitated by points. **B)** Palatal bone segments separated from the dental cast base for further analysis of the estimated volume.



The statistical analysis was performed by o GraphPad Prism software (Prism 5 for Windows - Version 5.0 – GraphPad software., Inc. San Diego, USA), with level of significance of 5%. Normality was checked by Shapiro-Wilk test. The methodological reliability was evaluated by measuring 1/3 of the sample twice, at 15-day interval. Paired t test evaluated the intraexaminer analysis, while independent t test assesses the interexaminer analysis. Dahlberg formula quantified the casual error. Repeated measures ANOVA followed by Tukey test and Friedman test followed by Dunn test was used for intragroup comparisons. Independent t test and Mann-Whitney test was used for intergroup comparisons. Data were presented as mean/standard deviation (SD) and median/interquartile amplitude (IA) in parametric and non-parametric analyses, respectively.

Results

Forty children were selected for the study. G1 (n=20) had 10 males and 10 females, while G2 had 14 males and 6 females. One hundred and twenty dental cast were analyzed. The mean age was 0.35 (± 0.12) years at T1, 1.14 (± 0.21) years at T2, and 2.08 (± 0.16) years at T3. No statistically significant differences occurred in the analysis of the intraexaminer (paired t test, $p = 0.244$, Dahlberg formula = 0.066) and interexaminer errors (independent t test, $p = 0.311$).

G1 showed a statistically significant volume increase after the 1st post-surgical phase (T2) followed by a statistically significant reduction at T3 (p=0.003). G2 exhibited a statistically significant volume increase between T1xT2 (p=0.001) (Table 1). Considering gender, the comparisons of all times revealed no statistically significant differences for both groups (Table 2).

Table 1 – Intragroup analyses of the estimated volumes (cm³) – ANOVA followed by Tukey test; Friedman test followed by Dunn test.

	T1		**T2**		**T3**		
	Mean (Median)	**SD (AI)**	**Mean (Median)**	**SD (AI)**	**Mean (Median)**	**SD (AI)**	***P***
G1	(2.79) A	(0.96)	(4.05) B	(1.76)	(2.72) A	(1.27)	**0.003*†**
G2	2.59 A	0.68	3.83 B	0.74	3.18 AB	1.37	**0.001***

SD: standard deviation. IA: Interquartile amplitude.
† Friedman test followed by Dunn test. * Statistically significant differences.
Different capital letters in line mean statistically significant difference.

Table 2 – Analyses of the estimated volume (cm³) according to gender (independent t test and Mann-Whitney test).

	Male		**Female**		
G1	**Mean (Median)**	**SD (IA)**	**Mean (Median)**	**SD (IA)**	***P***
T1	(2.40)	(1.02)	(2.93)	(0.38)	0.393‡
T2	3.94	1.80	4.50	1.21	0.432
T3	3.23	1.15	2.65	1.11	0.269
G2					
T1	(2.46)	(0.55)	(2.74)	(0.97)	0.063‡
T2	(3.87)	(0.27)	(3.34)	(1.13)	0.433‡
T3	(3.16)	(1.71)	(2.29)	(1.43)	0.201‡

SD: standard deviation. IA: Interquartile amplitude.‡ Mann-Whitney test.

No statistically significant differences occurred between groups, at all periods (T1, T2, and T3) (Table 3). The analyses of the volume differences (Δ = T2 – T1; T3 – T1; T3 – T2) revealed no significant differences between groups (Table 4).

Table 3 – Intergroup analyses of the estimated volumes (cm³) (Independent t and Mann-Whitney test).

	T1	T2	T3
P (G1 X G2)	0.640	0.315	0.542‡

‡ Mann-Whitney test.

Table 4 – Intergroup analyses of the estimated volume differences (∆;cm³) between times – independent t test.

	G1		G2		***P***
	Mean	**SD**	**Mean**	**SD**	
T2 – T1	1.52	1.55	1.24	1.21	0.562
T3 – T1	0.24	1.27	0.58	1.45	0.441
T3 – T2	-1.27	2.14	-0.65	1.51	0.292

SD: standard deviation.

Discussion

In this present study, we evaluate the palatal volume in children with UCLP, before and after two different surgical protocols through 3D digitized models. The literature lacks longitudinal studies on the use of digital resources to measure the palatal volume in children with oral clefts, at the first childhood, after the primary plastic surgeries. Thus, this study complements the literature on the analysis of the dental arches of children with oral clefts submitted to surgical approaches at the first months of life [5,11,18,17,20,21]. In this present study, the palatal growth after the two surgical protocols was restricted, with a transversal decrease. The transversal growth inhibition can be caused by intrinsic factors (severe maxillary growth) and/or iatrogenic factors (early palatal surgery, pressure around the tissues, or healing tissue in the maxillary segments), inhibiting the transverse palatal growth.

In this study, in G1, the 1st post-surgical phase (T2) did not influence on the growth of the dental arches because of the volume increase. However, at T3, the volume significantly reduced, that is, one-step palatoplasty negatively influenced on the dental arch growth. The literature lacks consensus on the hypothesis that palatoplasty accounts for the volume decrease of the dental arch of children with UCLP. In G2, the primary plastic surgeries did not impact on the dental arch growth, evidenced by the volume increase. At T3, the volume decrease, without statistically significant differences. The intergroup comparison of the estimated volume showed similarity between groups, at the evaluated periods.

The first study on the literature that measured the palatal volume was performed by Heiser et al., 2004[26], who indirectly measured the volume through the correlation with weight. In the study of Monga et al., 2020[27], the authors showed a statistically significant difference the palatal volume in individuals with UCLP compared to the controls and to individuals with bilateral cleft lip and palate. Ambrosio et al 2020[20], the intragroup volumetric analysis, after cheiloplasty (T2), both groups showed a significant growth, revealing that cheiloplasty itself did not impact on growth. This was similar to the results of Pucciarelli et al 2015[25], who also indicated a volume increase in children with UCLP after lip closure in children submitted to pre-cheiloplasty. The comparison with the results of this present study shows that after the lip closure, no change in the growth of the dental arches occur, that is, the volume remains unchanged.

One study, in 2017, evaluated the palatoplasty through von Langenbeck technique in individuals with oral clefts, while other [27] evaluated the flap palatoplasty technique. The results of this study confirmed the discovery of a long-term study with 25 years conducted by Michael Mars from 1984 to 2009. They found a greater reduction of the maxillary arch measured by Goslon scale in the individuals submitted to palatoplasty with Veau-Wardill-Kilner flap compared with the Oslo samples, in which the individuals underwent palatoplasty by von Langenbeck technique [27]. The result of this present study after the use of palatoplasty by von Langenbeck technique revealed a non-significant reduction in the volume. The literature lacks studies to support the hypothesis that palatoplasty accounts for the dental arch volume reduction in children with oral clefts. Notwithstanding, the study of Ambrosio et al 2020[20] found a reduction after palatoplasty (T3), corroborating the results of this present study.

A clinically relevant difference in the palatal volume in children with UCLP was observed in this study, indicating a greater maxillary constriction. The rationale behind this fact can be explained by three reasons. The first reason is the pressure after the lip reconstruction. The second is the constriction force on the palate due to healing tissue post-palatoplasty. And the third is a lack of arch continuity with the premaxilla protuberance, with is present in the cases of bilateral cleft lip and palate, resulting in the collapse of the lateral segments [27]. One of interesting findings of this present study is that the primary palatoplasty can also influenced on the severity of the maxillary growth restriction. Notwithstanding, further studies are necessary with homogeneous methodology and sample and to test the relationship with the airways. No consensus exists whether the disturb on the medium facial third occurs due to the cleft itself or is a consequence of the surgical techniques. Thus, the longitudinal analysis of the maxillary volume can contribute for this knowledge and improve the rehabilitation protocols.

Conclusion

The surgical protocol influenced the palatal volume of children with unilateral cleft lip and palate. Over time, in the same group, this study suggested that two-step palatoplasty was more suitable for dental arch development.

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4 GENERAL CONCLUSIONS

The surgical protocol influenced the severity of the maxillary growth restriction of children with unilateral cleft lip and palate.

The palatoplasty can influence in the palatal volume of children with unilateral cleft lip and palate.

Over time, in the same group, this study suggested that two-stage palatoplasty less affected the volume ofthe palate of children with unilateral cleft lip and palate.

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