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

Avaliação volumétrica do palato em crianças com fissura labiopalatina: comparação de dois protocolos cirúrgicos

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ABSTRACT

Objective: Compare the palatal volume in children with unilateral cleft lip and palate before and after two surgical protocols. **Material and Methods:** Retrospective data collection was performed in a specialized hospital. The sample comprised 120 digitized dental models divided into, Group 1 (G1) – participants submitted to cheiloplasty at 3 months (Millard technique) and one-step palatoplasty at 12 months (von Langenbeck technique); Group 2 (G2) – participants submitted to cheiloplasty (Millard technique) and hard palate closure (Hans Pichler technique) at 3 months and soft palate closure at 12 months (Sommerlad technique). The dental models were evaluated at Time 1 (T1): before primary plastic surgeries, Time 2 (T2): 1st post-surgical phase, and Time 3 (T3): 2nd post-surgical phase. The volume was measured through stereophotogrammetry system software. Parametric and non-parametric statistical tests were applied (α=5%). **Results:** 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 ($p=0.001$). There was no statistically significant difference in the intergroup and gender analyses ($p>0.05$). **Conclusion:** The surgical protocol influenced the palatal volume of children with unilateral cleft lip and palate. This study suggested that two-step palatoplasty protocol has a tendency to be more appropriate.

KEYWORDS

Cleft lip; Cleft palate; Dental arch; Imaging; Three-dimensional.

RESUMO

Objetivo: Comparar o volume palatino em crianças com fissura unilateral de lábio e palato antes e após dois protocolos cirúrgicos. **Material e Métodos:** A coleta de dados retrospectiva foi efetuada em um hospital especializado. A amostra foi composta por 120 modelos dentários digitalizados divididos em, Grupo 1 (G1) – participantes submetidos a queiloplastia aos 3 meses de vida (técnida de Millard) e a palatoplastia em única etapa aos 12 meses (técnica de von Langenbeck); Grupo 2 (G2) – participantes submetidos a queiloplastia (técnica de Millard) e fechamento do palato duro (técnica de Hans Pichler) aos 3 meses de vida e fechamento do palate mole aos 12 meses (técnica de Sommerlad). Os modelos dentários foram avaliados em Tempo 1 (T1): antes das cirurgias plásticas primárias, Tempo 2 (T2):1ª fase pós-cirúrgica e Tempo 3 (T3): 2ª fase pós-cirúrgico. O volume foi mensurado por meio do software do sistema de estereofotogrametria. Testes estatísticos paramétricos e não-paramétricos foram utilizados (α=5%). **Resultados:** As análises intragrupos indicaram que G1 apresentou aumento estatisticamente significante em T2 seguido de redução em T3 ($p=0.003$). G2 apresentou crescimento estatisticamente significativo do volume palatino entre T1 e T2 ($p=0.001$). Não houve diferença estatisticamente

significante nas análises intergrupos e entre gêneros (p>0.05). **Conclusão:** O protocolo cirúrgico influenciou o volume palatino das crianças com fissura unilateral de lábio e palato. Este estudo sugeriu que o protocolo da palatoplastia em duas etapas possui uma tendência de ser mais apropriado.

PALAVRAS-CHAVE

Fenda labial; Fissura palatina; Arco dentário; Imageamento tridimensional; Procedimentos cirúrgicos bucais.

INTRODUCTION

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

The rehabilitative protocol begins with the primary surgeries (cheiloplasty – lip repair surgery - and palatoplasty - palate repair surgery), which are aimed at restoring the face symmetry and correcting the anatomic defect, improving esthetics and function to enable favorable conditions and quality of life [5,6]. The Millard technique of cheiloplasty comprises the projection of relaxing incisions that enable the flap rotation to close the lip [7]. The most used technique for palatoplasty is the von Langenbeck technique, described in 1861. In this technique, relaxing incisions enable the displacement of mucoperiosteal flaps, that is sutured at nasal septum level, fixed only by the palatal vascular bundle [8]. In 1926, Hans Pichler included the vomer flap to close the hard palate. 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. Its repositioning aimed to restore the intravelar muscle, providing mobility to the palatal veil and improving the velopharyngeal competency [9]. Sommerlad technique, also called radical intravelar veloplasty, has shown excellent results for speech by reducing the velopharyngeal inadequacy [10].

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 performing 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 speech development [13]. Thus, the literature lacks consensus on primary surgery type, technique, and time that would result in smaller restrictive effects on the maxillary growth of these individuals [14-17].

The analysis of the dental molds through pre-determined anatomic points marked on three-dimensional (3D) images has 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 submitted to different surgical protocols. This study aimed at gathering knowledge on the aspects interfering on the maxillary development of children with unilateral cleft lip and palate (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 approved by the local Ethical Committee (CAAE: 77285417.0.0000.5417). Healthy children with UCLP, operated by the same surgeon, who did not begin the rehabilitative treatment participated in this study. Exclusion criteria were individuals with

other malformations and/or syndrome; absent documentation or poor-quality dental casts.

The sample size was obtained according to the study of Pucciarelli et al. [25], considering a standard deviation of 0.49 cubic centimeters (cm^3) for the greater bone segment before the lip closure, with $\alpha=5\%$, power analysis of 80%, and the minimal clinically important difference of 0.45 cm^3. Twenty children per group were the minimum sample size.

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

The participants had the impressions of the dental arch at three different periods, Time 1 (T1) – before primary plastic surgeries; Time 2 (T2) – 1st post-surgical phase; Time 3 (T3) – 2nd post-surgical phase. The dental casts were digitized by a surface scanning laser (Scanner R700™ Scanner; 3Shape AS, Copenhagen, Denmark). Two examiners analyzed the digitized dental arches through 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. [25], and quantified in cm^3. 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 was 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).

The statistical analysis was performed using GraphPad software Version 5.0 (Prism 5 for Windows., Inc. San Diego, USA), with $\alpha=5\%$. Normality was checked by the Shapiro-Wilk test. The methodological reliability was evaluated by measuring 1/3 of the sample twice, at a 15-day interval [20]. Paired t-test evaluated the intraexaminer analysis, while the unpaired 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. Unpaired t-test and Mann-Whitney test were used for intergroup comparisons. Data were presented as mean/standard deviation (SD) and median/interquartile range (IR) 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 (n=20) had 14 males and 6 females. One hundred and twenty dental molds were analyzed. The mean age was 0.35 (SD 0.12) years at T1, 1.14 (SD 0.21) years at T2, and 2.08 (SD 0.16) years at T3. No statistically significant differences occurred in the analysis of the intraexaminer (paired t-test, p = 0.244,

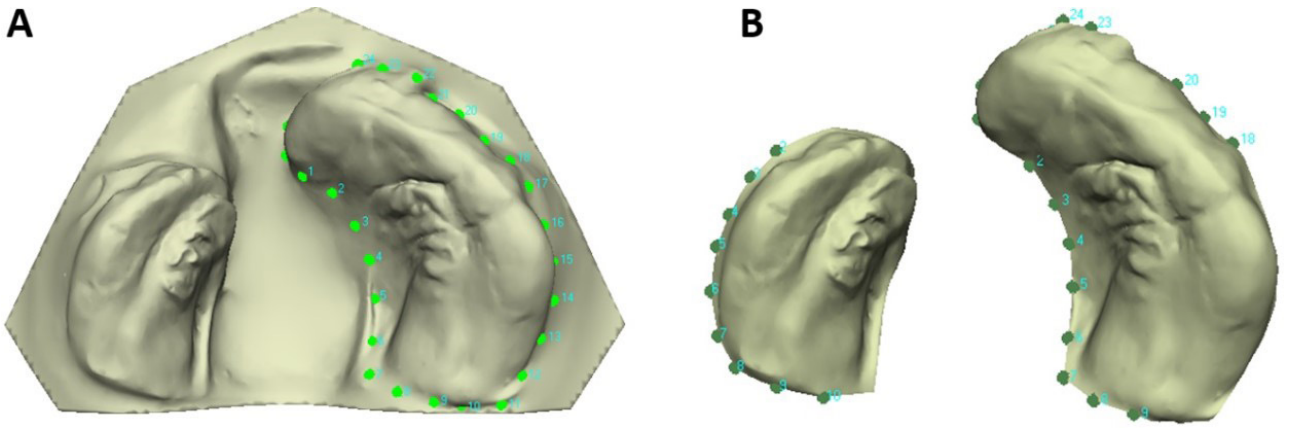

Figure 1 - Dental arch with unilateral cleft lip and palate. (A) Palatal bone segment delimitated by points; (B) Palatal bone segments separated from the dental cast base for further analysis of the estimated volume.

Dahlberg formula = 0.066) and interexaminer errors (unpaired 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 T1 *vs.* T2 (p=0.001) (Table I). Considering gender, the comparisons of all times revealed no statistically significant differences for both groups (Table II). No, statistically significant differences occurred between groups, at all periods (T1, T2, and T3) (Table III). The analyses of the volume differences (Δ = T2 – T1; T3 – T1; T3 – T2) revealed no significant differences between groups (Table IV).

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, in 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 [5,11,17,18,20,21]. In this present study, the palatal growth after the two surgical protocols was modified.

Table I - Intragroup analyses of the estimated volumes (cm^3) – ANOVA followed by Tukey test; Friedman test followed by Dunn test

	Time 1 Mean (Median)	SD (IR)	Time 2 Mean (Median)	SD (IR)	Time 3 Mean (Median)	SD (IR)	*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*

G1: Group 1; G2: Group 2. SD: Standard Deviation; IR: Interquartile Range. †Friedman test followed by Dunn test. *Statistically significant differences. Different capital letters in line mean statistically significant difference.

Table II - Analyses of the estimated volume (cm^3) according to gender – unpaired t-test and Mann-Whitney test

Sample	Male		Female		*P*
Group 1	Mean (Median)	SD (IR)	Mean (Median)	SD (IR)	
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
Group 2					
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‡

T1: Time 1; T2: Time 2; T3: Time 3. SD: Standard Deviation; IR: Interquartile Range.‡Mann-Whitney test.

Table III - Intergroup analyses of the estimated volumes (cm^3) – unpaired t-test and Mann-Whitney test

Group 1 *vs.* Group 2	Time 1	Time 2	Time 3
P	0.640	0.315	0.542‡

‡Mann-Whitney test.

Table IV - Intergroup analyses of the estimated volume (cm^3) differences (△) between times – unpaired t-test

Δ	Group 1 Mean	SD	Group 2 Mean	SD	*P*
T2 – T1	1.52	1.55	1.24	1.21	0.526
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. T2 – T1: Time 2 – Time 1; T3 – T1: Time 3 – Time 1; T3 – T2: Time 3 – Time 2.

In this study, in G1, the 1st post-surgical phase (T2) did not influence 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 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 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 statistically similarity between groups, at the evaluated periods. Thus, the different surgical protocols did not interfere in the dental arch volume.

The first study on the literature that measured the palatal volume was performed by Heiser et al. [26], who indirectly measured the volume through the correlation with weight. In the study of Monga et al. [27], the authors showed a statistically significant difference between the palatal volume in individuals with UCLP compared to the controls and individuals with bilateral cleft lip and palate. Ambrosio et al. [20], the intragroup volumetric analysis, after cheiloplasty, both groups showed significant growth, revealing that cheiloplasty itself did not impact on growth. This was similar to the results of Pucciarelli et al. [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 occurs, that is, the volume remains unchanged.

One study, in 2017, evaluated the palatoplasty through the von Langenbeck technique in individuals with oral clefts [28], 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 the 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. [20] found a reduction after palatoplasty, corroborating the results of this present study.

CONCLUSIONS

The surgical protocol influenced the palatal volume of children with unilateral cleft lip and palate. This study suggested that two-step palatoplasty protocol has a tendency to be more appropriate. Further studies should be carried out in these individuals, as before and after orthodontic treatment.

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Authors' Contributions

ECPA: Conceptualization, Methodology, Software, Validation, Formal Analysis, Writing – Original Draft Preparation and Writing – Review & Editing. ARHA: Conceptualization and Investigation. PKJ: Methodology, Software, Validation and Data Curation. CFCC: Investigation and Visualization. FPV: Formal Analysis and Writing – Review & Editing. SS: Writing – Original Draft Preparation and Writing – Review & Editing. MAAMM: Writing – Review & Editing. TMO: Conceptualization, Supervision, Project Administration, Writing – Review & Editing and Funding Acquisition.

Conflict of Interest

The authors have no proprietary, financial, or other personal interest of any nature or kind in any product, service, and/or company that is presented in this article.

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Regulatory Statement

This study was conducted in accordance with all the provisions of the local human subjects oversight committee guidelines and policies of the University of São Paulo, Bauru School of Dentistry. The approval code for this study is 77285417.0.0000.5417.

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