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Post-surgical effects on the maxillary segments of children with oral clefts: New three-dimensional anthropometric analysis

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Summary

This study aimed to use new three-dimensional (3D) anthropometric analyses to verify the post-surgical effects on the maxillary segments of children with unilateral cleft lip and palate. The sample was composed of digitized dental models of 60 children with unilateral complete cleft lip and alveolus (UCLA) and complete unilateral cleft lip and palate (UCLP). The impressions were taken before cheiloplasty (T1), after cheiloplasty (T2), and after palatoplasty (T3). The 3D anthropometric analyses of digitized dental casts were obtained through a specific software. Intragroup changes were applied paired t test and Wilcoxon test (UCLA group) and for the UCLP group, repeated-measures analyses of variance followed by the Tukey test. For intergroup analyses, an independent t test and Mann-Whitney test were used. The palatal dimensional changes of UCLA group showed that the distances I–C, I–T’, and I–T significantly increased after cheiloplasty (p=0.0002, p=0.0007 and p<0.0001, respectively). In the UCLP group, the I–C’ distance statistically decreased in the post-surgical periods (p<0.0001), while the I–T distance increased (p<0.0001). The I–C distance increased after cheiloplasty (p<0.0001). The I–T’ distance increased between T2 and T3 with statistically significant differences (p=0.0037). The intergroup analysis of palatal development (T2-T1) showed that the distances I–C’ and I–T’ demonstrated a reduction of the dental arches growth of UCLP group compared with the UCLA group, with statistically significant differences. The new 3D anthropometric analysis showed that the development of the maxillary segments changed after surgical repair. The UCLP group demonstrated a reduction of the dental arches growth compared with the UCLA group.
Keywords: cleft lip, cleft palate, imaging, three-dimensional, anthropometry, surgery, plastic.
INTRODUCTION

In dentistry, researchers use digital anthropometry to analyze the dental arch development of children with cleft lip and palate (CLP) undergoing reparative plastic surgeries such as cheiloplasty and palatoplasty (Sakoda et al., 2017; Falzoni et al., 2016; Jorge et al., 2016). These surgical procedures are indispensable methods for the anatomic and functional rehabilitation performed usually at 3 months (cheiloplasty) and 12 months (palatoplasty) of life (Freitas et al., 2012).

Surgery improves the physiological and psychological aspects of these children; however, the maxilla development is influenced not only by the characteristics of the congenital defects (Chiu et al., 2011; Zhang et al., 2015), but also by the surgical procedures carried out in early childhood (Falzoni et al., 2016; Shi, Losee, 2015; Zhang et al., 2015). The evidence of changed maxillary growth could be analyzed through dental casts with the benefit of performing a longitudinal following-up of the rehabilitative protocol (Fernandes et al., 2015) without exposure to ionizing radiation.

The early analysis of palatal growth enables verifying how each cleft type behave after the surgical procedures (Sakoda et al., 2017) and can suggest the surgical technique and time most indicated for the correction of each cleft type, thus modifying rehabilitative protocols (Fernandes et al., 2015). This would ensure more appropriate growth, and consequently, harmony between esthetic and functional factors. Therefore, this study aimed to use new 3D anthropometric analyses to verify the post-surgical effects on the maxillary segments in children with unilateral cleft lip and palate.

MATERIAL AND METHODS
This study was submitted to and approved by the Institutional Review Board regarding the ethical aspects. A total of 150 dental casts were obtained through the files of the Hospital for the Rehabilitation of Craniofacial Anomalies, University of São Paulo, Brazil (HRAC/USP). The rehabilitation protocol regarding lip repair was performed with Millard’s technique around 3 months of age. Complete palate repair was performed with Von Langenback’s technique around 12 months. Inclusion criteria were children of either sex with unilateral complete cleft lip and alveolus (UCLA) and unilateral complete cleft lip and palate (UCLP). Exclusion criteria were children with syndromes or those without complete dental documentation.

Sample size calculation considered the study of Harila et al. (2013) with a standard deviation of 1.83 mm. Considering the level of significance of 5%, test power of 80%, and the minimum difference to be clinically detected of 1.4 mm, the minimum sample size was 28 children. Thus, the study sample comprised 30 children with UCLA (12 boys and 18 girls) and 30 children with UCLP (17 boys and 13 girls). The dental casts of each child were obtained at the following periods: T1, before cheiloplasty (UCLA and UCLP groups); T2, after cheiloplasty (UCLA and UCLP groups); and T3, after palatoplasty (UCLP group).

The dental casts were digitized (Scanner 3Shape R700 Scanner, Copenhagen, Denmark) (Sakoda et al., 2017; Falzoni et al., 2016; Jorge et al., 2016) and the anthropometric analyses were performed by the software of a stereophotogrammetry system (Mirror Imaging Software, Canfield Scientific Inc., Fairfield, NJ, USA) in the Laboratory of Functional Anatomy of the Stomatognathic System, University of Milan, Italy (Céron-Zapata et al., 2016; De Menezes et al., 2016). Anatomic landmarks and anthropometric measurements were: I–C (anterior inter-segment distance: interincisor point to the point of eruption of the primary canine of the greater segment); I–T
(anterior-posterior inter-segment distance: interincisor point to tuber of the greater segment); I–C’ (anterior intra-segment distance: interincisor point to the point of eruption of the primary canine of the lesser segment); and I–T’ (anterior-posterior intra-segment distance: interincisor point to tuber of the lesser segment) (Figure 1 A – B). All measurements were performed by a trained and calibrated examiner as in previous studies (Falzoni et al., 2016; Jorge et al., 2016; Sakoda et al., 2017; Fuchigami et al., 2017; Shetty et al., 2017).

All statistical analyses were performed with GraphPad Prism software (Prism 5 for Windows, version 5.0; GraphPad Software, Inc.) with a level of significance of 5%. The intra-examiner error was analyzed through repeated-measures analysis 15 days after the first measurements in one-third of the sample, randomly selected. To analyze the systematic and casual error, a paired t test and Dahlberg’s formula were respectively used. Data distributions were verified for all variables; for normally distributed values, means and standard deviations were calculated, and inferential parametric tests were used. Otherwise, medians, interquartile amplitudes and non-parametric tests were used. To verify the intragroup changes in the UCLA group, a paired t test and Wilcoxon test were applied. In the UCLP group, repeated-measures analysis of variance followed by the Tukey test were applied. The intergroup comparisons used an independent t test and the Mann-Whitney test.

**RESULTS**

The median ages (in years) of the children were verified at all study periods. In UCLA group, the median ages were 0.29 and 1.74 respectively at T1 and T2. The median ages of UCLP group were 0.29 (T1), 1.08 (T2), and 2.25 (T3). To assess
reproducibility, the intra-examiner error was analyzed, and showed no statistically significant differences in the repeated-measures analysis (p>0.05).

The palatal dimensional changes of the UCLA group showed that the distances I–C, I–T’, and I–T significantly increased after cheiloplasty (Table 1). In the UCLP group, the I–C’ distance statistically decreased in the post-surgical period, while the I–T distance increased. The I–C distance increased after cheiloplasty, with statistically significant differences. The I–T’ distance increased between T2 and T3 with statistically significant differences (Table 2). Table 3 displays the intergroup analysis of palatal development (T2-T1) and shows that the distances I–C’ and I–T’ demonstrated a reduction of the dental arch growth in the UCLP group compared with the UCLA group, with statistically significant differences (Table 3).

**DISCUSSION**

Currently, digital anthropometric analysis is a viable alternative to conduct studies to verify the development of the dental arches in children with cleft lip and palate undergoing reparative surgical procedures (Carrara et al., 2016; Falzoni et al., 2016; Sakoda et al., 2017). The 3D measurements can verify the differences between children with and without congenital orofacial anomalies (Fernandes et al., 2015), analyze how different cleft types develop after the same surgical procedures (Sakoda et al., 2017), compare rehabilitative protocols (Jorge et al., 2016), and assess the growth of the dental arches in children with pre-surgical orthopedics (Céron-Zapata et al., 2016; Fuchigami et al., 2017; Shetty et al., 2017).

The present study analyzed the post-surgical effects on the development of the maxillary segments of children with non-syndromic CLP who did not undergo pre-surgical orthopedics. All children had cheiloplasty performed at 3 months of age.
(Millard technique) and one-stage palatoplasty at 12 months (von Langenbeck technique) in HRAC/USP.

In the UCLA group, the I–C’ distance was greater before surgery than the I–C distance, but at T2, the I–C’ distance showed inhibition, which strengthens the hypothesis that dental arches in children with oral clefts grow under the influence of the reparative surgeries (Shi, Losee, 2015; Zhang et al., 2015; Falzoni et al., 2016) and of the cleft size (Chiu et al., 2011; Zhang et al., 2015). Thus, a directly proportional relationship occurs between the anatomic cleft size and the post-surgical dysmorphic growth.

In the UCLP group, the anthropometry of the anterior palatal area (I–C and I–C’) demonstrated that on average the anterior inter-segment distance (I–C’) reduced after all post-surgical periods, while the I–C distance significantly increased after cheiloplasty (T2) and remained stable after palatoplasty (T3). Concerning the anterior-posterior distances, the I–T’ distance increased only between T2 and T3, while the I–T distance increased after all study periods. Several studies found comparable results in the assessment of the anterior palatal area through the intercanine distance. After cheiloplasty, the intercanine distance significantly reduced in children with UCLP (Falzoni et al., 2016; Jorge et al., 2016; Sakoda et al., 2017). Heliövaara et al. (2017) reported that regardless of the therapeutic approach, medical professionals always seek good dentofacial development of individuals with cleft lip and palate. However, a consistent finding in the scientific literature is the collapse of the maxillary dental arches propitiating a retrusion of the maxilla, which can cause modifications dental arch relationships. At T3, I–T and I–T’ distances did not decrease, although the I–C’ distance decreased and the I–C distance seemed to be inhibited. Some authors believe that the palatoplasty can inhibit the sagittal development of the maxilla (Tome et al., 2016). Indeed, it is difficult to determine whether palatoplasty can inhibit anterior palatal growth or whether the
inhibition continues due to the iatrogenic effect of early performed cheiloplasty. According to Huang et al. (2002), cheiloplasty can exert an uninterrupted pressure through the scar tissue on the anterior palatal area, and further studies are necessary to quantify the post-surgical lip pressure on the dental arch.

The changes in the maxillary development of children with CLP can also be three-dimensionally analyzed through facial morphology (Dádákova et al., 2016; Wu et al., 2016). The facial analysis conducted by Dádákova et al. (2016) identified few differences in facial symmetry in children with and without CLP. Moreover, the analysis demonstrated that facial growth in the children with CLP was changed in comparison with that in children without clefts, probably because of the cleft itself and the need of palatoplasty (Dádákova et al., 2016).

The literature lacks consensus on which would be the reparative surgery with more iatrogenic effects on the development of dental arches. This lack of consensus may rely on other factors that may influence growth, such as the following: the size of the anatomic defect (Chiu et al., 2011); the genetic pattern of the craniofacial growth of each individual (Honda et al., 1995); the presence of syndromes or associated anomalies, and the different surgical techniques and periods (Shi, Losee; 2015); the use of pre-surgical orthopedics (Céron-Zapata et al., 2016; Fuchigami et al., 2017; Shetty et al., 2017); and the surgeon’s ability (Stancheva et al., 2015).

Thus, further quantitative analyses of the development of the dental arches (angulations, volume of the palatal bone segments, perimeter, and superposition of the dental arches) are necessary to quantify the differences existing among the different cleft types, from birth to skeletal maturity. This will enable the tailoring of an individualized rehabilitative protocol for each type of orofacial anomaly that favors a correct and harmonic maxillary growth.
CONCLUSION

The new 3D anthropometric analysis showed that the development of the maxillary segments changed after the repair surgeries. The UCLP group demonstrated a reduction of the dental arches growth compared with the UCLA group.
Conflict of interest

The authors report no conflict of interest.

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Figure 1. (A, B) Anatomic points of dental arches. (A) Dental arch of unilateral complete cleft lip and alveolus (UCLA). (B) Dental arch of unilateral complete cleft lip and palate (UCLP).
Table 1. Palatal dimensional changes (mm) in UCLA group, at T1 and T2 (paired t test and Wilcoxon test).

<table>
<thead>
<tr>
<th>Variable</th>
<th>T1 Mean (median)</th>
<th>T1 SD (IA)</th>
<th>T2 Mean (median)</th>
<th>T2 SD (IA)</th>
<th>P</th>
</tr>
</thead>
<tbody>
<tr>
<td>I–C’</td>
<td>17.27</td>
<td>2.05</td>
<td>17.88</td>
<td>1.84</td>
<td>0.096</td>
</tr>
<tr>
<td>I–C</td>
<td>14.03</td>
<td>2.08&lt;sup&gt;a&lt;/sup&gt;</td>
<td>15.41&lt;sup&gt;a&lt;/sup&gt;</td>
<td>1.51&lt;sup&gt;a&lt;/sup&gt;</td>
<td>0.0002&lt;sup&gt;*&lt;/sup&gt;</td>
</tr>
<tr>
<td>I–T’</td>
<td>36.39</td>
<td>2.34</td>
<td>38.15</td>
<td>2.62</td>
<td>0.0007&lt;sup&gt;*&lt;/sup&gt;</td>
</tr>
<tr>
<td>I–T</td>
<td>33.19</td>
<td>2.68</td>
<td>35.80</td>
<td>2.5</td>
<td>&lt;0.0001&lt;sup&gt;*&lt;/sup&gt;</td>
</tr>
</tbody>
</table>

IA, interquartile amplitude; SD, standard deviation; UCLA, unilateral complete cleft lip and alveolus.

<sup>a</sup> Median and IA (interquartile amplitude), Wilcoxon test.

<sup>*</sup>Statistically significant difference.
Table 2. Palatal dimensional changes (mm) in UCLP group, at T1, T2, and T3 (analysis of variance followed by Tukey test).

<table>
<thead>
<tr>
<th>Variables</th>
<th>T1 Mean</th>
<th>T1 SD</th>
<th>T2 Mean</th>
<th>T2 SD</th>
<th>T3 Mean</th>
<th>T3 SD</th>
<th>P</th>
</tr>
</thead>
<tbody>
<tr>
<td>I – C’</td>
<td>18.81</td>
<td>3.43</td>
<td>14.98</td>
<td>2.92</td>
<td>13.34</td>
<td>2.65</td>
<td>&lt;0.000*</td>
</tr>
<tr>
<td>I – C</td>
<td>13.03</td>
<td>1.59</td>
<td>15.38</td>
<td>1.46</td>
<td>15.99</td>
<td>1.38</td>
<td>&lt;0.000*</td>
</tr>
<tr>
<td>I – T’</td>
<td>35.89</td>
<td>2.90</td>
<td>35.13</td>
<td>2.61</td>
<td>36.54</td>
<td>2.83</td>
<td>0.0037*</td>
</tr>
<tr>
<td>I – T</td>
<td>30.21</td>
<td>2.37</td>
<td>33.24</td>
<td>2.31</td>
<td>35.11</td>
<td>2.58</td>
<td>&lt;0.000*</td>
</tr>
</tbody>
</table>

SD, standard deviation; UCLP, unilateral complete cleft lip and palate.

* Statistically significant difference. Equal lowercase letters in line denote statistically significant differences.
Table 3. Intergroup analysis of the palatal development (mm) at T2–T1 (independent $t$ test and Mann-Whitney test).

<table>
<thead>
<tr>
<th>Variable</th>
<th>UCLA Mean (median)</th>
<th>UCLA SD (IA)</th>
<th>UCLP Mean (median)</th>
<th>UCLP SD (IA)</th>
<th>$P$</th>
</tr>
</thead>
<tbody>
<tr>
<td>I–C'</td>
<td>0.60</td>
<td>1.94</td>
<td>-3.83</td>
<td>2.52</td>
<td>&lt;0.0001*</td>
</tr>
<tr>
<td>I–C</td>
<td>1.76 $^A$</td>
<td>2.17 $^A$</td>
<td>2.50 $^A$</td>
<td>2.14 $^A$</td>
<td>0.160</td>
</tr>
<tr>
<td>I–T'</td>
<td>1.75</td>
<td>2.56</td>
<td>-0.75</td>
<td>2.26</td>
<td>0.0002*</td>
</tr>
<tr>
<td>I–T</td>
<td>2.61</td>
<td>2.93</td>
<td>3.03</td>
<td>2.84</td>
<td>0.572</td>
</tr>
</tbody>
</table>

IA, interquartile amplitude; SD, standard deviation; UCLA, unilateral complete cleft lip and alveolus; UCLP, unilateral complete cleft lip and palate.

*Median and IA, Mann-Whitney test.

*Statistically significant difference.