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Palatal growth in complete unilateral cleft lip and palate patients following neonatal cheiloplasty: Classic and geometric morphometric assessment

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A B S T R A C T

Background: A new method of early neonatal cheiloplasty has recently been employed on patients having complete unilateral cleft lip and palate (cUCLP). We aimed to investigate (1) their detailed palatal morphology before surgery and growth during the 10 months after neonatal cheiloplasty, (2) the growth of eight dimensions of the maxilla in these patients, (3) the development of these dimensions compared with published data on noncleft controls and on cUCLP patients operated using later operation protocol (LOP; 6 months of age).
Methods: Sixty-six virtual dental models of 33 longitudinally evaluated cUCLP patients were analysed using metric analysis, a dense correspondence model, and multivariate statistics. We compared the palatal surfaces before neonatal cheiloplasty (mean age, 4 days) and before palatoplasty (mean age, 10 months).
Results: The palatal form variability of 10-month-old children was considerably reduced during the observed period thanks to their undisturbed growth, that is, the palate underwent the same growth changes following neonatal cheiloplasty. A detailed colour-coded map identified the most marked growth at the anterior and posterior ends of both segments. The maxilla of cUCLP patients after neonatal cheiloplasty had a growth tendency similar to noncleft controls (unlike LOP).
Conclusions: Both methodological approaches showed that early neonatal cheiloplasty in cUCLP patients did not prevent forward growth of the upper jaw segments and did not reduce either the length or width of the maxilla during the first 10 months of life.

1. Introduction

Orofacial clefts are among the most common craniofacial deformities [1–3] and are associated with serious orthodontic anomalies [4]. The incidence in the Czech Republic is approximately 1:530 of living newborns. Their background is multifactorial, in other words, orofacial clefts are caused by a combination of genetic and environmental factors [5]. They are the result of hypoplasia of facial prominences and palatal shelves and growth restriction of the mandible [6]. This malformation emerges from approximately the fourth to the eighth week of prenatal development. The range of the affliction varies, being localised to the lip, upper jaw, and palate, separately or in different combinations [1]. Complete unilateral cleft lip and palate (cUCLP), the subject of our study, is the most common type of orofacial cleft [7,8].

Treatment of cleft patients should begin as soon as possible [9] and includes surgical repair of the cleft lip, cleft palate, affected nose, along with orthodontic therapy [7]. The surgical treatment goals are mainly to restore the form and function of structures affected by clefting [10] and thus improve facial appearance and, ultimately, influence the psychological impact on the child and

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family [9]. The associated treatment goals are to improve speech and food intake [11].

Primary cheiloplasty is necessary to reconstruct the normal anatomy and function of the lip [12], and the most common age for surgical treatment is between 3 and 6 months [13,14]. Cheiloplasty can also be performed during the first week of life, and it is becoming the most common surgical approach in the Czech Republic. Neonatal cheiloplasty is believed to result in many benefits such as excellent wound healing, feeding facilitation [9,11,13], and reducing the psychological impact on family [15]. The question is whether neonatal cheiloplasty results in a better or the same outcome as the later operation protocol (LOP). We evaluated the isolated influence of neonatal cheiloplasty on maxillary growth during the first 10 months after lip surgery. We compared maxillary morphology before and 10 months after neonatal cheiloplasty using classical morphometry combined with three dimensional (3-D) geometric morphometric methods. The aim of our study was to determine whether neonatal cheiloplasty has any negative effect on the growth of the maxillary segments during the observed period. We compared our morphometric data with published data on noncleft controls and cUCLP patients operated using the LOP to prove our hypothesis.

2. Materials and methods

This study was based on morphometric analysis of plaster models of the maxillae of 33 patients with cUCLP. All the patients were of Czech origin and were operated within the first week of life by the same surgeon using the modified Tennison technique. The consent for experimentation with human subjects was obtained. The mean age of the 33 patients who underwent early neonatal cheiloplasty was 3.8 ± 2.7 days, and the mean age for palatoplasty was 10.1 ± 1.8 months. Two plaster casts were taken of each patient, the first before cheiloplasty (T0) and the second before palatoplasty (T1). The plaster casts were scanned using a Breuckmann SmartScan scanner (Aicon 3D Systems GmbH, Braunschweig, Germany) with a resolution of 0.1 mm. The resulting meshes were edited and decimated using RadpidForm XOS software (Inus Technology Inc, Seoul, South Korea).

The first step before any morphometric analysis was to place 11 landmarks on each model in Morphome3cs software (www.morphome3cs.com; Fig. 1). Landmark placement error was determined according to the method of von Cramon-Taubadel et al. (2007) [16], at 0.1665 mm. We used those 11 landmarks to analyse changes in seven linear measurements and one angular measurement during the first 10 months: 1–7 (alveolar cleft width), 5–9 (intermolar width), 4–8 (intercanine width), 4–6 (molar region length on the noncleft side), 8–10 (molar region length on the cleft side), 6–10 (intertuberosity width), 2–11 (palatal length), and ∠1-3-7 (anterior basal angle). Some of the measured dimensions were compared with published data of a noncleft control group [17] and with an LOP group (cheiloplasty at 6 months of age) [18]. We used Wilcoxon's paired signed-rank test to determine whether growth in the specified dimensions was statistically significant. Significance was decided to be at level $\alpha = .05$.

We then analysed the palatal shapes in their entirety using coherent point drift dense correspondence analysis (CPD-DCA) [19]. This is an algorithm that transfers the topology of one surface, called the base, to all the other surfaces. In effect, homologous samplings of these surfaces are generated, allowing for landmark-style statistics and visualisations. CPD-DCA first uses an automatic nonrigid registration algorithm to deform the base mesh onto each nonbase surface, bringing anatomically similar locations into close proximity. Next, closest-point search is used to find corresponding points on the nonbase surface, which are used to resample the nonbase surface. These homologous samplings of each surface are then aligned with generalised Procrustes analysis (GPA) [20]. Note that size was not normalised during GPA and, as a result, the real form of the individuals was analysed. Dimension reduction is accomplished using principal component analysis (PCA); the shape variables reduced to the first two principal components were plotted in a scatter plot. The mean growth direction in the space of the first two principal components was calculated as, where n is the number of specimen pairs and $\boldsymbol{b_i}$ and $\mathbf{a}_i$ are the principal component (PC) scores of the i-th individual after and before surgery, respectively.

Finally, mean shapes for pre- and postsurgery patients were calculated using means of their corresponding vertex coordinates. Average growth pattern was calculated as the average corresponding vertex displacement between pre- and postsurgery surfaces. This mean displacement was visualised by colour-coding the groupwise mean surfaces.

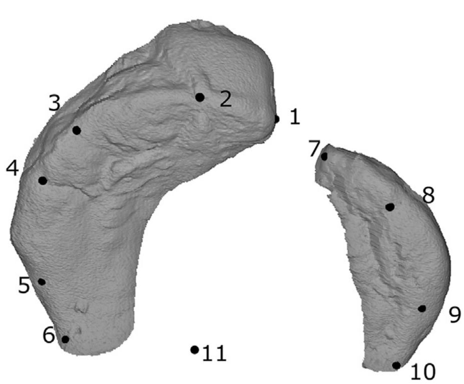

Fig. 1. Reference points on maxillary segments. 1: Most distal point on the edge of the segment on noncleft side; 2: tip of ridge on the line between the labial frenulum and incisive papilla; 3: mesial margin of canine swelling on noncleft side; 4: distal margin of canine swelling on noncleft side; 5: distal margin of molar swelling on noncleft side; 6: tuberosity point on noncleft side; 7: most mesial point on the edge of the alveolar segment on cleft side; 8: distal margin of canine swelling on cleft side; 9: distal margin of molar swelling on cleft side; 10: tuberosity point on cleft side; 11: reference point on the base of the perpendicular line from reference point 2 to the line segment of reference points 6 and 10.

3. Results

3.1. Classical metric analysis

Seven linear and one angular dimension of the maxilla of cUCLP patients were compared between two age groups (T0, T1), one at age 3.8 days and the second at age 10.1 months (Table 1). All the measured dimensions were compared by Wilcoxon signed-rank test. Table 1 shows significant growth increments in all the measured dimensions except intercanine width (4–8), whose size change was not statistically significant. There was an evident decrease in the dimensions associated with convergence of the anterior ends of the upper jaw segments such as cleft width (1–7) and anterior basal angle (1-3-7). On the contrary, there was a significant growth change in the posterior area of upper jaw segments: increase of intertuberosity width (6–10), intermolar width (5–9), and molar region length on the cleft side (8–10) and noncleft side (4–6). There was also a significant growth extension in palatal length (2–11).

A further important aim of our study was a metric comparison (intercanine width, intertuberosity width, and palatal length) of our data with noncleft controls [17] and with another UCLP-patient group operated using a classical LOP [18] (Fig. 2). Initial