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Three-dimensional development of the upper dental arch in unilateral cleft lip and palate patients after early neonatal cheiloplasty

E. Hoffmannova[a,*], V. Moslerová[a,b], J. Dupej[a,c], J. Borský[d], Š. Bejdová[a], J. Velemínská[a]

[a] Department of Anthropology and Human Genetics, Faculty of Science, Charles University, Prague, Vinicna 7, 128 00 Prague 2, Czech Republic
[b] Department of Biology and Medical Genetics, 2nd Faculty of Medicine, Charles University and Motol University Hospital, V Úvalu 84, 150 00 Prague 5, Czech Republic
[c] Department of Software and Computer Science, Faculty of Mathematics and Physics, Charles University in Prague, Czech Republic
[d] Department of Otorhinolaryngology 2nd Faculty of Medicine, Charles University, Prague, Faculty Hospital Motol, V Úvalu 84, 150 00 Prague 5, Czech Republic

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ABSTRACT

Objectives: This prospective morphometric study evaluated the growth of the upper dental arch in UCLP patients after early neonatal cheiloplasty and compared the selected dimensions with published data on non-cleft controls and on later operation protocol patients.
Methods: The sample comprised 36 Czech children with nonsyndromic complete UCLP (cUCLP) and 20 Czech children with nonsyndromic incomplete UCLP (UCLP + b). 2-D and 3-D analyses of palatal casts were made at two time points: before neonatal cheiloplasty at the mean age of 3 days (± 1 day), and 10 months after surgery at the mean age of 10 months (± 1 month).
Results: The upper dental arch of cUCLP and UCLP + b patients showed similar developmental changes, but the cleft type influenced growth significantly. The initial high shape variability in cUCLP patients diminished after 10 months, and approached the variability in UCLP + b patients. Both the width and length dimensions increased after surgery. Important growth concerned the anterior ends of both segments. The width and length dimensions illustrated similar growth trends with non-cleft controls and UCLP patients who underwent later cheiloplasty.
Conclusion: Early neonatal cheiloplasty caused no reduction in the length or width dimensions during the first year of life. Our data suggest a reconstructed lip has a natural formative effect on the actively growing anterior parts of upper dental arch segments, which cause narrowing of the alveolar cleft.

1. Introduction

Orofacial clefts are associated with serious developmental anomalies of both hard and soft tissues [1–4]. The growth disturbance of the maxilla is typical in UCLP patients [5] and is suspected to result from congenital hypoplastia, and growth deficiency caused by scar formation resulting from lip or palatal repair [6].

Treatment of cleft patients starts with surgical repair of the cleft lip and/or palate with special attention to the soft tissues of the lip and nose [7,8]. The repair of the lip physiologically restores the continuity of the upper lip musculature [9]. Nowadays, cheiloplasty is performed more and more often in neonates during the first week of life, but widely accepted timing for surgical treatment is still between 3 and 6 months of age [10,11]. Such neonatal cheiloplasty solves the problems resulting from the cleft lip as soon as possible. The essential motive for proposing neonatal surgery is the psychosocial impact on mother – baby interaction and other family members [11–13]. Benefits of neonatal cheiloplasty include outstanding wound healing with barely visible scars, facililiation of feeding, child socialisation, and dealing with OME (otitis media effusion) [9,10,12,14,15]. It is argued that the later operation protocol (LOP) – performed between 3 and 6 months of age – yields better aesthetic results since the postnatal increase in size of anatomic structures allows their easier accessibility and manipulation during surgery [16], but early neonatal cheiloplasty performed by an experienced surgeon has similar aesthetic and orthodontic results [17].

Treatment outcome and favourable or unfavourable growth of upper dental arch can be also anticipated according to the initial severity of the cleft deformity [18] where patients having a small alveolar cleft develop a more protruded (i.e., less hypoplastic) maxilla than those with a large alveolar cleft [19].

A new method (a modification of the method of Tennison) has recently been employed for surgical repair of unilateral cleft lip in newborns with UCLP operated during the first week of life [9]. The

* Corresponding author. Tel.: +420 732 11 86 49.
E-mail addresses: eva.hoffmannova87@gmail.com, ewulk@seznam.cz (E. Hoffmannova).

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modification consists of using two tissue flaps from the margins of the cleft lip to deepen the oral vestibule (VF) and floor of the nasal vestibule and to bring extra tissue to the suture region (NF).The Tennison's flap was used to complete the shortened philtrum on the cleft side. This provides a suture with minimum tension of the lip tissue after surgery. The orbicularis oris muscle was isolated and nasal septum was repositioned. The rotated and dislocated nasal alar cartilage on the cleft side was released and its position and shape corrected. .A more detailed description of neonatal cheiloplasty was published in our previous report [14].

The aim of our current study is to evaluate and compare growth of the upper dental arch in patients with different cleft types before and after neonatal cheiloplasty using two-dimensional (2-D) and three-dimensional (3-D) methods.

2. Patients and methods

This prospective study is part of a long-term research project focused on the influence of neonatal cheiloplasty on growth and development of the face. The study was based on morphometric analysis of plaster casts of the upper dental arch of 56 patients of Czech origin with nonsyndromic UCLP: 36 of the patients had complete UCLP (cUCLP) and 20 patients had UCLP with either a soft or a combined tissue bridge (UCLP + b). The soft and combined tissue bridge groups were grouped together, since no significant differences between the groups were found. The persisting tissue bridge is located at the lower margin of the nostril and soft tissue and skeletal bridge across the anterior maxilla. The soft tissue adhesions connect the divided maxillary fragments in the absence of the bony union of the maxillae at the cleft side.

Two plaster casts were available for each patient, one taken before cheiloplasty at the mean age of 3 days (± 1 day) - age 0, the other at the mean age of 10 months (± 1 month) - age 1. The plaster casts were made under anesthesia. The surgery was performed by the same surgeon within the first week of life.

The plaster casts were scanned using a Breuckmann SmartScan scanner (Aicon 3D Systems GmbH, Braunschweig, Germany) with a resolution of 0.1 mm. The meshes were edited using RapidForm XOS software (INUS Technology, Inc, Seoul, Korea).

Morphometric analysis was based on 11 landmarks that were placed on each model according to a modified approach developed by Mazaheri et al. [20] (Fig. 1). Furthermore, six linear measurements and

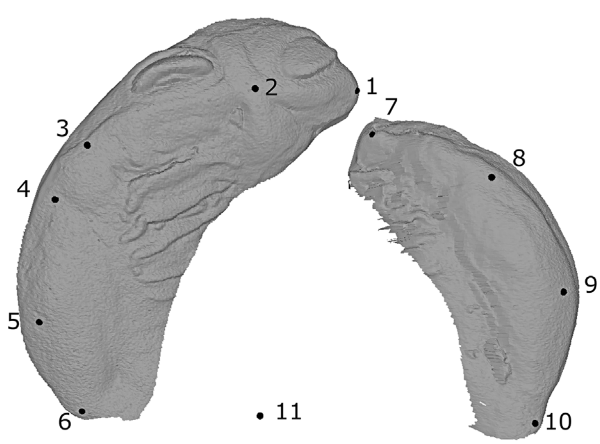

Fig. 1. Reference points on the alveolar arch. 1- the most distal point on the edge of the alveolar segment on the noncleft side; 2 - tip of the ridge on the line between labial frenulum and incisive papilla; 3 - mesial margin of canine swelling on the noncleft side; 4 - distal margin of canine swelling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

one angular measurement were taken: 1–7 (alveolar cleft width), 5–9 (inter-molar 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), ∠1–3 – 7 (anterior basal angle), and 2–11 (palatal length). Landmark placement and all further processing was performed using Morphome3cs software (www.morphome3cs.com). Landmark placement error was determined [21] at 0.15 mm.

In order to evaluate changes in size during the studied period, the differences in measurements and the post-treatment changes in dimensions between cleft types (cUCLP and UCLP + b) were tested using Student's t-test. Statistical significance was decided at $\alpha = 0.05$.

Both cUCLP and UCLP + b groups were further subdivided by cleft severity inferred from cleft width at age 0, into three subgroups: mild (0–5 mm, N = 22), moderate (5–10 mm, N = 16), and severe (> 10 mm, N = 18). Two-way analysis of variance (ANOVA) was used to verify the effect of cleft type (cUCLP or UCLP + b) and cleft severity (mild, moderate, severe) on the measurements. We also tested the interaction between the two factors. The differences between the specified measurements before and after treatment in both cleft groups were used as the dependent variable.

Selected dimensions were compared to those of non-cleft control groups and UCLP patients operated following the LOP (3–6 months of age). The control group data and the data of the later cheiloplasty groups were taken from Kramer et al. [22].

In order to perform statistical analysis on the surface data, vertex homology among the meshes had to be created; the following outlines the applied procedure. We used dense correspondence analysis (DCA), described by Hutton et al. [23], which transfers the topology of an arbitrarily-selected base mesh to all other meshes. In effect, all meshes were resampled in a way that creates homologous vertices, which can be treated as landmarks in further processing; therefore, they are dubbed quasilandmarks. Note, that before further processing, all surfaces were aligned to a common mean, using generalized Procrustes analysis (GPA) on their landmarks.

Principal component analysis (PCA) was performed on the quasilandmark coordinate matrix to reduce the dimensions of the data, while simultaneously minimizing the loss of information. The transformed specimens and confidence ellipses denoting group variability for both cleft types and ages were visualized in a PCA scatter plot.

Taking advantage of the availability of two plaster casts for each patient (taken in the first week and at 10 months of age), we modelled the longitudinal growth of the upper dental arch in each cleft type (cUCLP and UCLP + b) during the first 10 months after early neonatal cheiloplasty. The registered vertex coordinates of pre- and post-treatment stages were subtracted, which produced the growth displacement fields. The mean growth was visualized as a colour map along with pre- and post-treatment mean surfaces. Colour encodes the distance travelled by each particular feature during growth, with blue denoting little growth, and red indicating extensive growth.

3. Results

3.1. Shape variability

Shape variability of the upper dental arch in patients with cUCLP and UCLP + b was analysed by PCA of the respective shape variables. Their scores in the first two principal components (PC1, PC2) are showen in a scatter plot (Fig. 2). The shape variation explained by PC1 is 28.8% and 10.7%. by PC2. Each specimen appears twice in the plot: first, at age 0 and second, at age 1. The shape variability of the more serious cleft type (cUCLP) group at age 0, is notably larger than in the other three groups. The shape difference between cUCLP and UCLP + b at age 0 is mostly explained by PC1. Ten months after surgery, the shape variability of both cleft types decreased considerably. PC1 shows the decrease in alveolar cleft width and in shape variability of the upper dental arch, both of which are more apparent in the cUCLP group than