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Three-dimensional evaluation of facial morphology in pre-school cleft patients following neonatal cheiloplasty

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ABSTRACT

Purpose: To evaluate the facial morphology of pre-school patients with various types of orofacial cleft after neonatal cheiloplasty in pre-school aged children; and to compare facial variability and mean shape with age-corresponding healthy controls.
Materials and methods: The sample included 40 patients with unilateral cleft lip (CL), 22 patients with unilateral cleft lip and palate (UCLP), and 10 patients with bilateral cleft lip and palate (BCLP). Patients were divided into two age categories, with a mean age of 3 years and 4.5 years, respectively. The group of healthy age-matched controls contained 60 individuals. Three-dimensional virtual facial models were evaluated using geometric morphometry and multivariate statistics methods.
Results: Statistically significant differences were found between each of the cleft groups and the controls. Color-coded maps showed facial shape deviations, which were located mainly in the nasal area and philtrum in all groups examined, and also in the buccal region and the chin in patients with UCLP or BCLP. These differences became more apparent, but not significantly so, in the older age category.
Conclusion: Facial deviations typical of patients with clefts were observed in all of the patient groups examined. Although the analysis showed statistically significant differences in overall facial shape between patients and controls among all groups tested, the facial morphology in patients who have undergone only neonatal cheiloplasty (CL) is influenced to a small extent and may be considered satisfactory. More severe cleft types (UCLP, BCLP) together with palatoplasty, are reflected in more marked impairments in facial shape.

1. Introduction

Orofacial clefts are among the most common types of congenital structural malformations worldwide. The incidence varies from 1/500 to 1/2500 cases, depending on geographic location and ethnicity (Schutte and Murray, 1999; Vanderas, 1987). In persons of white ethnicity, the figure is between 1/600 and 1/700 (Murray, 2002; Tolarova and Cervenka, 1998; Vlastos et al., 2009).

During normal craniofacial growth, the oronasal components fuse together and form facial structures including the palate and lip (Weinberg, 1994). Any alteration during the process can disrupt or prevent the closure of components at the right time, and thus result in clefting. Consequently, the skeletal and tissue growth of the whole area is affected negatively (Diah et al., 2007; Weinberg, 1994).

Primary surgical closure of the soft tissue of the lip and nose (i.e., primary cheiloplasty) is the first and probably the most discussed step in the treatment of patients with a cleft lip or cleft lip and palate (Habel et al., 1996; Schutte and Murray, 1999). The main aims of primary surgical treatment are the restoration of orofacial tissue, which enables normal functioning such as feeding, and reconstruction of facial appearance (Borsky et al., 2012). If the defect is

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more severe or is combined with cleft palate, other surgeries usually follow (Habel et al., 1996).

The worldwide accepted timing range for performing primary cheiloplasty is between 2 and 6 months of age; it is usually done at 3 months of age (Harris et al., 2010; Mcheik et al., 2006). In the past 20 years, the idea of performing surgery at an alternative time, early in the neonatal period (i.e., within approximately 10 days of birth) has emerged. It has been suggested that a successful surgery performed earlier in life can minimize the negative impact (especially psychosocial) of the cleft (Goodacre et al., 2004).

The advantages and disadvantages of such early surgery are often discussed in the literature. The original concerns were related especially to the safety of general anesthesia at such an early age. However, no difference was found in the probability of cardiac arrest or occurrence of perioperative complications between neonates and 3-month-old children (Bhananker et al., 2007; Murat et al., 2004).

Moreover, it has been suggested that healing and scar formation are different shortly after birth than at later ages (Li et al., 2014). During intrauterine development, instead of scarring, fetal fibroblasts deposit matrix in a pattern that resembles normal skin (Adzick and Lorenz, 1994). This process, called scarless healing, can persist even after birth for a short period of time. Thus, an earlier timing of surgery can be beneficial for healing and also for the overall appearance of the cleft area (Adzick and Longaker, 1992; Borsky et al., 2012). The aesthetic results of the surgery are as good as in patients undergoing delayed surgery. This implies that surgeons are equally able to achieve a favorable outcome regardless of the timing of the repair (Goodacre et al., 2004).

Psychosocial advantages speak in favor of neonatal timing as well. Parents prefer to bring their neonate home with no visible deformity. Immediate closure of the cleft also simplifies feeding of the infant and thus, later, it can even have a positive impact on weight gain and normal growth (Borsky et al., 2012).

Overall, the current literature mostly agrees on the benefits of neonatally performed cheiloplasty (Akin et al., 1991; Goodacre et al., 2004; Harris et al., 2010; Borsky et al., 2012; Petráčková et al., 2015). Despite this, not much has been written about growth and development of the face itself after the procedure and in later years. The initial results suggest that the data acquired on maxillary growth after neonatal surgery (Borsky et al., 2012) are consistent with findings obtained on a group of patients operated on at an average age of 3.2 months (Huang et al., 2002). According to Akin (Akin et al., 1991), neonatal surgery has a significantly positive occlusive effect on the alveolar arch, which does not need further surgical correction.

This study aimed to evaluate facial morphology in various preschool age patients with orofacial cleft who underwent neonatal cheiloplasty, in comparison with healthy controls of corresponding ages.

2. Materials and methods

This study was approved by the Ethics Committee of The Institutional Review Board of Charles University, Faculty of Science. The study was performed in accordance with the guidelines of the Declaration of Helsinki. Declaration and informed consent forms were signed by parents/legal representatives of the children before any intervention.

The participants in this study included patients with unilateral cleft lip (CL), unilateral cleft lip and palate (UCLP), and bilateral cleft lip and palate (BCLP). Individuals with atypical clefts or associated syndromes were not included. All patients underwent surgery at the Faculty Hospital Motol, Prague, Czech Republic, by the same surgeon. Primary cheiloplasty was performed using the same modified Tennison method within the first 8 days (or, exceptionally, 14 days) of life. Palatoplasty in the UCLP and BCLP groups was performed at a mean age of 10.21 months using the Furlow technique. The cleft patients studied consisted of two separate age categories, a younger category with a mean age of 3 years (2.5–3.7 years) and an older category with a mean age of 4.5 years (4.0–5.0 years); both categories contained subgroups with each of the three cleft types. The younger age category consisted of 42 patients (26 CL, 12 UCLP, 4 BCLP) and the older age category 30 patients (14 CL, 10 UCLP, 6 BCLP).

A control group of healthy children, from preschools in Letná and Hrabákova in Prague, were in the same age range as the patient group; the younger category comprised 22 individuals, the older 38 individuals.

Methods of geometric morphometry applied to three-dimensional (3D) facial scans allowed us to evaluate the facial surface in its entirety and to assess even minor changes precisely.

For the analysis, 3D facial models were used. Images were acquired by a noninvasive optical technique, using a high-resolution Vectra 3D scanner (Canfield Scientific, Inc., Fairfield, NJ, USA). During capture, each participant was seated in front of the scanner with the head in a natural position; the patient was asked to make a neutral facial expression. The final 3D facial model was generated using the associated software Mirror PhotoTools (Canfield Scientific, Inc.). Afterwards, each model was imported into the software RapidForm 2006 (INUS Technology Inc., Seoul, Korea) for further processing, which involved manual trimming of unwanted data or noise, filling holes in the model, and decimation.

Before statistical processing, vertex homology had to be enforced in the entire sample of surfaces. To that end, we used CPD-DCA (Dupej et al., n.d.), a modification of the original dense correspondence analysis (DCA) algorithm by Hutton et al. (2001). In contrast to DCA, which relies on landmark-fitted spatial deformations, CPD-DCA uses an automatic nonrigid registration algorithm coherent point drift (CPD) to find corresponding vertices, which results in more accurate correspondences outside the convex hull of the landmarks.

The processing started with rigid registration of the surfaces. Nine landmarks (exoR = right exocanthion; exoL = left exocanthion; enR = right endocanthion; enL = left endocanthion; N = nasion; Pn = pronasale; chR = right cheilion; chL = left cheilion; Pg = pogonion) were manually placed on every model, according to Hutton et al. (2001). Generalized Procrustes analysis (GPA) was performed on these landmarks and the resulting transformations were used to rigidly align the entire meshes. Next, a template mesh, called a base, was arbitrarily selected from the sample, as reported by Hutton et al. (2001). As long as the mesh had an even coverage of vertices, its choice had negligible bearing on the subsequent statistics. Then, the base was registered to each non-base mesh (called floating) with an automatic nonrigid algorithm. After registration, the closest points to the deformed base were found on the floating mesh, and these were considered homologous to the base's vertices. Because of this assumed homology, these vertices are dubbed quasi-landmarks. Poor correspondences were detected by ascertaining triangle compaction, and such quasi-landmarks were removed from further processing.

Finally, principal components analysis (PCA) was performed on the vertex coordinate matrix to reduce data dimensionality. Quasi-landmarks that were improperly matched were removed from the PCA and therefore did not introduce any unwanted variability. For purposes of visualization, these quasi-landmarks were imputed with a thin plate spline-based approach (Dupej et al., n.d.). This entire process was performed using the software suite Morphome3cs (http://www.morphome3cs.com/).