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3-D shape analysis of palatal surface in patients with unilateral complete cleft lip and palate

Hana Rusková [a], Šárka Bejdová [a], Miroslav Peterka [b], Václav Krajíček [a,c], Jana Velemínská [a,*]

[a] Department of Anthropology and Human Genetics (Head: Assoc. Prof. Vladimír Sládek, M.Sc., Ph.D.), Faculty of Science, Charles University in Prague, Viničná 7, 128 43 Prague 2, Czech Republic

[b] Institute of Experimental Medicine Academy of Sciences CR (Head: Prof. Eva Syková, M.D., D.Sc.), Prague, Czech Republic

[c] Department of Software and Computer Science Education (Head: Assoc. Prof. Dr. Pavel Töpfer, Ph.D.), Faculty of Mathematics and Physics, Charles University in Prague, Prague, Czech Republic

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A B S T R A C T

Facial development of patients with unilateral complete cleft lip and palate (UCLP) is associated with many problems including deformity of the palate. The aim of this study was to evaluate palatal morphology and variability in patients with UCLP compared with Czech norms using methods of geometric morphometrics. The study was based on virtual dental cast analysis of 29 UCLP patients and 29 control individuals at the age of 15 years.

The variability of palatal shape in UCLP patients was greater than that in nonclefted palates. Only 24% of clefted palates fell within the variability of controls. The palatal form of UCLP patients (range from 11.8 to 17.2 years) was not correlated with age.

Compared with control palates, palates of UCLP patients were narrower, more anteriorly than posteriorly. Apart from the praemaxilla region, they were also shallower, and the difference increased posteriorly. The UCLP palate was characterised by the asymmetry of its vault. The maximum height of the palatal vault was anterior on the clefted side, whereas it was posterior on the nonclefted side. The slope of the UCLP palate was more inclined compared with the control group. The praemaxilla was therefore situated more inferiorly.

1. Introduction

Orofacial clefts in newborns are some of the most common facial deformities; their causes are multifactorial. They are generated by hypoplasia of facial prominences and palatal shelves and growth retardation of the mandible in human (O'Rahilly and Müller, 1992), as well as by experimental models (Jelínek and Peterka, 1977; Peterka and Jelínek, 1983).

The incidence of the orofacial clefts is about 1–7 per 1000 liveborn babies. The children with orofacial clefts need multidisciplinary care from birth to adulthood (e.g. Dürwald and Dannhauer, 2007; Vlastos et al., 2009). The presence of malformation has a long-lasting adverse outcomes for health and social integration. The communication of the cleft patients is affected because of their problems with speech and hearing. The later timing of hard palate closure significantly affects speech development (e.g. Krey et al., 2009; Mossey et al., 2009).

Unilateral complete cleft lip and palate (UCLP), the subject of our study, is the most frequent type of orofacial malformation. For these individuals, growth disturbance of the maxilla and the large number of orthodontic anomalies is typical (Jelínek et al., 1983). In order to choose the correct surgical technique and the timing of the surgery and start of orthodontic treatment, it is essential to know the growth potential of the patient's jaws (Peterka and Pěnkava, 1975).

For individuals with UCLP, decreased growth of the maxilla anteriorly is typical. In addition to reduced palatal growth, posteriorotation of the mandible often develops. Significant deterioration in intermaxillary relationships occurs mainly during puberty in relation to the eruption of permanent teeth. Limited pubertal acceleration with significant growth of the mandible is typical in UCLP individuals (Šmahel and Brejcha, 1983). Increase in alveolar arch is important for the correct placement of teeth, which is also limited in this case. Hypoplasia of the upper jaw can also lead to

* Corresponding author. Tel.: +420 221 951 612; fax: +420 221 951 619.
E-mail address: velemins@natur.cuni.cz (J. Velemínská).

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anterior crossbite and to palatal or vestibular eruption of teeth (Peterka et al., 1980).

Later development of the upper jaw is the result of sutural growth, bone remodelling, growth of the alveolar ridges and eruption of the teeth (Enlow and Hans, 1996). In early postnatal development, round arch forms change into oval arch forms (Sillman, 1964; Kramer et al., 1992). The dental arch of 3-year-old children with UCLP or bilateral cleft lip and palate (BCLP) before surgery is wider than the unaffected dental arch. This is caused by unconnected segments of the alveolar arch (Jelínek et al., 1983). Indeed, measurements are significantly larger in unoperated adults with BCLP than in controls because of the unmerged parts of the palate (Diah et al., 2007). The antero-posterior dimension of the alveolar arch in UCLP, BCLP and cleft palate patients is shorter (Jelínek et al., 1983). An important feature of UCLP individuals is that their palate surface is smaller than in healthy subjects (Huddart et al., 1978; Lo et al., 2003), which suggests that there is an intrinsic tissue deficiency in the palate/maxilla before palatoplasty (Lo et al., 2003). After lip surgery, the dental arch narrows (Huang et al., 2002).

A rapid reduction of palatal width was found after palatoplasty, which patients undergo from 3 to 6 years of age. Also, diminished palatal length has been described during all the evaluated periods from 3 to 18 years (Peterka et al., 1980). According to Schliephake et al. (2006), asymmetry (cleft side vs. noncleft side) of the anterior part of the palate in UCLP patients is greater than that of the molar area, and it increases with age. In another study, UCLP patients in at 15 years were characterised by a certain asymmetry compared with the control group palates (Šmahel et al., 2004).

The palate as a whole was shallower than in controls (Kilpeläinen et al., 1996; Šmahel et al., 2004). All maxillary dental arch dimensions in UCLP patients at ages 3–12 were smaller than in controls, except for intermolar width at age 12 (Athanasiou et al., 1988). The width between permanent first molars in 12-year-old UCLP patients was close to that of controls (Peterka et al., 1980). At age 15, dental arch width was narrower across all palatal surfaces. Arch length was assessed only between the first molars, and there was no reduction compared with controls (Šmahel et al., 2004).

To evaluate the shape of the palate in the above-mentioned studies, traditional morphometry was used. These studies often suffer from magnification errors, with consequent loss of information from the third dimension (Bugaighis et al., 2010).

Previous research of Šmahel et al. (2004) was based on evaluation of more than 200 dimensions, but it did not evaluate palatal shape variability or detailed praemaxilla description. From a methodological point of view, our study was built on the research of Bejdová et al. (2012), wherein the shape of the palate was evaluated in BCLP patients. Shape analysis, including the dense correspondence algorithm (DCA; Hutton et al., 2001), principal component analysis (PCA) and finite element scaling analysis (FESA), made it possible to evaluate the surface of the palate as a whole, its average changes and its interindividual variability.

The aim of this study was to use geometric morphometric tools to evaluate the variability and average differences of palatal shape in patients with UCLP compared with those in healthy Czech children.

2. Material and methods

2.1. Patients and control group

We analysed two groups consisting of 58 plaster casts of the maxillary alveolar arch and palate.

The UCLP sample consisted of 29 plaster casts of Czech boys. The casts were made as a part of the treatment record. Eighteen clefts were left sides and 11 clefts were of the right. Average age of the patients was 14.8 years (range from 11.8 to 17.2). All surgery was performed at the Clinic of Plastic Surgery in Prague from 1975 to 1980 by the same method. Lip surgery was performed according to Tennison, with primary periosteoplasty at an average age of 8.5 months (range, 5–17 months). Palatoplasty by pushback with two flaps and pharyngeal flap surgery was performed on patients at average age 4.9 years (range from 3.0 to 5.9). After surgery, the patients underwent long-term orthodontic treatment (Kuderová et al., 1996). Šmahel et al. (2004) have evaluated the treatment in detail. In our study, only UCLP patients without a bony bridge were included. Other conditions were absence of imperfections in the dental cast surface and at least partial eruption of both permanent second molars.

The control group consisted of plaster casts of 29 healthy boys of Czech origin. The average age was 14.7 years (range from 14.1 to 15.7). The casts had been taken in the 1970s in schools in the city of Brno. Conditions for inclusion in the study were at least partial eruption of both permanent second molars and absence of orthodontic anomalies (rotation, tooth position in the alveolar arch, crowding). Only individuals with an ideal occlusion were involved.

2.2. Methods

The maxillary plaster casts were digitised using laser scanner Roland LPX-250 (Roland DG, Hamamatsu, Japan) with a lateral resolution of 200 mm. The casts were scanned perpendicular to the occlusal plane using Dr. Picza 3 software (Roland DG), and the raw scanned data were processed using Pixform reverse engineering software (Roland DG). This software was used to revise the raw scans by removing the teeth from the models before analysis, because only the shape of the palate was evaluated (Fig. 1). It is well known that majority of the UCLP is localized on the left side. Our sample of UCLP patients comprised both left- and right-sided clefts. In order to have homogenized cleft side for further 3D shape analysis, the scans of right-sided clefts were flipped horizontally and evaluated as left-sided clefts.

The models were processed using the software MorphoStudio 3.02 and geometric morphometrics. DCA was used to convert the models to the same number of triangular faces (Hutton et al., 2001). In the first step, a set of four hand-placed landmarks was situated on each evaluated palatal surface. The first landmark was placed between the central incisors at the incisive papilla. The second and third landmarks were situated on the most medial cervical margins of the second premolars. The fourth landmark was placed in the middle of a line connecting the first molars at the most medial cervical margins between the mesiodistal and distopalatal cusps (more details in Bejdová et al., 2012).

The next step was the Procrustes superimposition (Zelditch et al., 2004), which was used to find the average landmark configuration of all four landmarks. After that, the models were transformed by thin-plate spline interpolation so that their landmark configurations fitted the average landmark configuration. All points or vertices of the polygon mesh can be further considered as landmarks and as the initial data for multivariate analysis.

Shape variability of clefted and nonclefted palates was analysed using principal component analysis (PCA). This method was suitable for studying individual variability as well as for comparing between groups of patients and controls, for which PCA score plots are suitable. Using the software mentioned above, we were able to reconstruct the palatal shape of any individual in the sample or along the axis of any principal component score. This methodology has frequently been used to evaluate facial variability (e.g., Singh et al., 2007; Weinberg et al., 2009).

As the age range in our UCLP sample was wide (from 11.8 to 17.2 years), linear regression was used to describe the relation between