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Dental arch morphology in adult cleft patients

Abstract: Using geometric morphometric methods to compare the dental arches of adult cleft patients with a unilateral cleft lip and palate to a group of eugnathic adults.

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Introduction

The development of the maxilla in patients with CLP is influenced by intrinsic and functional factors and the effects of cleft surgery (1). A tendency towards cranial movement in the smaller segment can be observed in surgically untreated adults with clefts (15 unilateral CLP). Patients with a bilateral cleft exhibit a constriction of the maxillary segments and protrusion of the premaxilla (2). The morphology of this group of patients also reveals significant differences in the severity of the cleft (3). Thus patients with unilateral clefts have a more V-shaped upper jaw with reduced anterior transverse width (4). The present results suggest that no detectable growth reduction (5) can be observed in patients with untreated clefts.
After surgical correction, characteristic changes in skeletal morphology and dental arches are to be expected (6, 7, 8).
Reduced growth in the transverse and sagittal direction (9) was already observed in childhood, though variation in individuals remains high.

In patients with unilateral clefts, **Johnson et al. (10]** was unable to find a correlation between the severity of the cleft at birth and conditions at the age of six. Thus, the study of adult patients seems justified regardless of the initial severity of the cleft.
A number of methods have been developed as part of the systematic study of the morphology of the maxilla in cleft patients. The contours drawn on the model were made accessible by **Stöckli (11)** through photocopying; **Berkowitz et al. (12)** used stereophotogrammetry for this, **Börnert et al. (13)** took a three-dimensional measurement with a reflex microscope and **Braumann et al. (14) used** a 3D laser scanner.
The most common approach is to obtain three-dimensional coordinates of defined points and by calculating distances and angles. The application of geometric morphometrics in this context could not be found in literature. The advantage of this coordinate-based procedure is that parameters which describe the size of a structure can be identified that do not depend on shape (**15**).
Below we will examine to what extent, in adulthood, specific differences in shape are still detectable despite orthodontic treatment and compare this with the eugnathic group.

Question
Do dental arches and apical bases differ in size and shape between the comparison group (noncleft) and adult patients with unilateral cleft lip and palate (cleft) who received late palate closure and orthodontic treatment?

Patients and Methods

Patients
Our study looked at the dental casts of 27 patients (> 18 years) with nonsyndromic unilateral CLP, who received late palate closure treatment (16, 17). The patients received no preoperative orthodontic treatment (Hotz plates). The lip was closed between the ages of 4 to 6 months according to **Millard (18);** primary osteoplasty and palatoplasty followed at the age of 4 years.
None of the patients received palatal expansion with a hyrax appliance or any kind of orthognathic surgery. Active orthodontic treatment included expansion plates for the maxilla in accordance with the methods of A.M. Schwarz as well as multi-bracket appliances. Only patients with no missing teeth (except for wisdom teeth) were selected. In some cases, lateral hypoplastic incisors were present in the cleft area. Four patients had clefts on the right side, 22 patients (85%) on the left side.
The cleft patients (17 men and 10 women, mean age 20.7 years, SD 8.05 years) were compared to a group of eugnathic adults (17 men and 10 women, mean age 24.5 years, SD 1.79 years). The dental casts came from the archives of the Department of Orthodontics from untreated adults with class I occlusion.

Data acquisition and landmarks
The survey was carried out using a three-dimensional Microscribe 3DX digitizer (Immersion Corp., San Jose, CA, USA). This is a passive robot arm with four degrees of freedom. It uses sensors to determine the exact position of the tip of the stylus in the space (xyz-coordinates). The accuracy rate is specified by the manufacturer with a sample rate of 1000 points/second at 0009 "in a workspace of 50".

Hayasaki et al. (19) already demonstrated the successful and precise measurement of model casts using this technology. In a methodological study, **Chen et al. (20)** extensively studied the suitability of the MicroScribe 3DX for measuring dental casts. **Penin-Lambert et al. (21)** also used this instrument in their three-dimensional analysis of mandibular morphology.
For the present study, we selected a combination of landmarks in accordance with **Korkhaus (22)** and, to calculate the dimensions of 1-12 (Dim1 - Dim12), we used landmarks chosen by **Sillman (23)** as well as some morphological points (**Fig. 1**). The measured distances were used to describe the transverse and sagittal Sillman-proportions of the dental arch. They were chosen because these data points and lines can be measured in a homologous way in all phases of development of dentition and therefore the data can also be used in further investigations.
In addition to the maxillary mesio-buccal cusps of the molars, we also scanned the buccal cusps of the premolars, the canines and the mesial and distal boundaries of the cutting edges of the incisors.

The landmarks for the description of the apical base were established by **Miethke et al. (24)**. The points are 5 mm below the apical point of the gingival margin of the lateral incisors, the canines and second premolars. This is a modification of the method proposed by **Rees (25).** The overjet and overbite were additionally measured with a digital caliper (Mannesmann, Mannesmann brothers Tools GmbH, Remscheid, Germany).

Statistics
We used the Shapiro Wilk test to check for normal distribution. The limit for the rejection of H0 (normal distribution) was set at $p \leq 0.05$. In the case of normal distribution, differences between groups were tested using the t-test for independent samples; otherwise the Wilcoxon-Mann-Whitney rank sum test (U test) was used. $p \leq 0.05$ was established as being statistically significant (*). The calculations were performed with R 2.12.2 (http://cran.r-project.org/).
Geometric morphometrics
The xyz coordinates were transferred to the program Paleontological Statistics (PAST) 2.07 (**26**; http://folk.uio.no./ohammer/past/) for morphological analysis.
The information on size was calculated as the normalized centroid size. It is a measurement that describes the size of an object regardless of its shape. Then shape-defining components could be identified using Procrustes transformation (**15**).
The centroid size (CS) univariate measurement does not depend on the position and orientation of the landmarks and is calculated directly from the coordinates. In accordance with **Dryden and Mardia (27, 28**) it is calculated as the square root of the sum of the squares of the distances of all points from the centroid. The somewhat more robust normalized centroid size used here is determined by dividing by the square root of the number of landmarks (15, **26**).
For the Procrustes transformation it is assumed that an object consists of a finite number of points *k* in *n* dimensions. By mathematically removing the components for translation, rotation and scaling, the object can be described by a set of equivalent classes (**29**). This process is repeated iteratively until the least square fit of all of the configurations cannot be improved upon further. Several objects with homologous landmarks can now be compared independent of their size (28, 29, **30, 31**).
Since only a limited number of models per study group was available, the number of measuring points had to be reduced to the specified number, even if more would have been

preferable for obtaining a better description. For the geometric morphometric studies, however, the number of landmarks should be smaller than the number of individuals studied (**15**).
The coordinates of the dental casts with right-side clefts were mirrored.

Methods of error
The error of the method in all three dimensions was calculated as described by **Dahlberg** (**32**). To do this, ten randomly selected models were digitized twice in a four week time interval.

Results

Error analysis
The calculated error was between 0.1 mm (premolar cusps) and 0.4 mm (cutting edge points of the incisors) in all dimensions. Only the tuberosity points showed errors up to 0.5 mm.

Traditional morphometrics
A significant difference between the groups could be found with respect to the incisors. The following measurements were observed for overjet and overbite:

$overjet_{cleft}$ = -1.3 mm; SD = 3.21 and $overjet_{noncleft}$ = 2.1 mm; SD = 0.74, p = 0.000
$overbite_{cleft}$ = 0.9 mm, SD = 2.27 and $overbite_{noncleft}$ = 2.9 mm; SD = 1.24, p = 0.000

Significant differences in the lower jaw could only be detected for the dimensions 3, 6, 8 and 9. These are to be regarded as a measurement of the length of the dental arch, which means there was a reduction in this dimension in the group of cleft patients. The widths of the dental arches in the anterior and posterior area (posterior arch width: PAW; anterior arch width: AAW) do not differ (Table I).
The values for the upper jaw revealed a completely different picture (Table II). The arch widths were significantly lower for the cleft group than for the control group.
The dimensions according to **Sillmann** (23) were significantly smaller in the group of cleft patients. Only dimensions 9 and 12 showed no significant differences between the groups. The anterior-posterior length of the dental arch and the distance between the tuberosity points (Dim 12) revealed no differences. In the case of a large standard deviation, the Dim 12 was the only value which increased.
In summary, the group of cleft patients showed significantly higher standard deviations in all distances measured.

Geometric morphometrics
Size
The following values were calculated for the dental landmarks of the lower jaw: CS_{cleft} = 23.58 and $CS_{noncleft}$ = 23.83 for the centroid size. CS_{cleft} = 27.55 and $CS_{noncleft}$ = 25.93 were calculated for the upper jaw.
As the hypothesis of normal distribution had to be rejected in the Shapiro Wilk test of the upper jaw (p_{cleft} = 0.0198 and $p_{noncleft}$ = 0.0491), testing was performed using a Wilcoxon-Mann-Whitney test (p = 0.4186). Data for the lower jaw could be tested for independent samples in the presence of normal distribution using the t-test (p = 0.3794). Hence there are no differences in size between the dental arches of the upper and lower jaw.

Centroid size calculated from landmarks of the apical base of the mandible showed no significant difference (CS_{cleft} = 18.3 and $CS_{noncleft}$ = 18.5, p = 0.548). The apical base of the maxilla in the cleft group was significantly smaller (CS_{cleft}= 20.7 and $CS_{noncleft}$ = 24.5, p = 0.000). The non-parametric Wilcoxon-Mann-Whitney rank sum test (U test) was performed because there was no normal distribution.

Shape

After Procrustes transformation it was possible to compare the two study groups in terms of the differences in shape (**Fig. 2**).

Only marginal differences in the shape of the mandible were visually evident and there was indication of a minimal flattening of the anterior arch. A significant constriction (cleft side) can be seen on the left side of the upper jaw. On the non-cleft side, the points of the canines and premolars are more buccal and anterior while on the cleft side there is a more distal positioning of the landmarks from the premolars onwards and a shift in the palatal direction. The second molars are subject to a visible shift buccally while the tuberosity points go in a palatal direction. The shape of the arch in the upper jaw ($p < 0.0001$) differs significantly between the cleft and noncleft groups. No differences could be found for the lower jaw, (p = 0.9854; ANOSIM of Procrustes residuals, Bray-Curtis distance, Bonferroni-corrected p).

Discussion

Error

The calculated error in the range of 0.1-0.5 mm is somewhat lower than what is found in the literature (**33**), but it appears that the identification of measurement points on fully toothed adults is more precise than on edentulous infant casts (**34**).

The relatively large standard deviations reflect the highly individual nature of the deviations in cleft patients. As **Honda et al. (9)** noted, this is not only explained by the operations. They found for example Dim 12 ± 3.19 mm for deviations, which is very approximate to what was measured here.

Traditional morphometrics

Overbite and overjet argue for a slightly open-mesial jaw relation in the group of cleft patients. As a camouflage therapeutic approach was pursued in these cases, a functionally and aesthetically satisfactory result can be expected upon successful completion of the treatment. The observed results, however, are less favorable than those found **by Gaggl et al. (35)**. The observed dental configuration is also consistent with the skeletal parameters of these patients measured in previous studies (**36**).

Mapes et al. 1(37) were able to show that in two to six year old patients with unilateral clefts, surgery-related growth retardation, at least in terms of the arch length, is made up for. This could explain the observed occurrence of Dim 9 (upper arch) in addition to the effects of orthodontic treatment.

The transverse deficits can be explained by reduced transversal growth as discovered by **Dahl (38)**. **Gaggl et al. (35)** also found sagittal and transverse underdevelopment of the maxilla. An expansion and extension in terms of post-development through purely orthodontic therapy may not be permanently stable (**39**) or possible to the extent deemed sufficient.

The shortened dental mandibular arch length could be explained as being the result of camouflage treatment with lingual-tipping of the incisors. The lower transverse width for bilateral clefts in the mandible, as described earlier by **Heidbüchel and Kuijpers-Jagtman**

(40), could not be found.

Geometric morphometrics
Even after orthodontic treatment in childhood and adolescence, the cleft patients we studied demonstrated characteristic changes in the shape of the upper jaw. The deficits found in the anterior area of the arch were already described in children by **Lentrodt et al. (41)** and **Dürwald and Dannhauer (17**).
The reduced overjet and reduced arch widths are consistent with studies conducted by **Marcusson** and **Paulin (42)**. Furthermore, an almost inevitable recurrence after transverse multi-bracket treatment (**39**) is expected. One reason may lie in the reduced basal sagittal and vertical development of the maxilla (**43**).
After orthodontic treatment, no size differences were found in the dental arches (centroid size). The orthodontic development was successful purely in terms of dentition; the development of the apical base was not adequately possible as revealed by the measurements. This may be caused by a tissue deficit in the anterior region as described previously (**44**).
The posterior enlargement in the area of the second molar was not immediately apparent using the methods of classic morphometics. In the literature this is attributed more to double-sided clefts (**45**). One reason may lie in therapeutic transverse expansion which contributes to a slightly posterior over-development. The transverse relapse described in this region (**46**) could not be confirmed. A vertical deviation in the small cleft segment could be established as before. The characteristics of the upper jaw shape are consistent with the anterior movement of the dentition on the non-cleft side, anterior-lateral displacements and midline deviations, all of which are familiar from clinical experience.
The observed characteristic shape of the arches and the transverse dimension could also be affected by changes after orthodontic treatment (**42**). These do not depend on the nature of the retention.
The surgical procedure of cleft closure is also an important aspect. According to **Gaggl et al. (47)** greater transverse and sagittal deficits can be expected after the sequential closure. There is a greater imbalance in the growth of the entire mid-face in these patients.
Furthermore, it has been shown that geometric morphometric methods are well suited for adequately describing changes when it comes to craniofacial deformities. This is also consistent with the results of other authors (**48, 49, 50**).

Conclusions
Using orthodontic therapy in conjunction with late palate closure, the size of the dental arches in patients with unilateral cleft lip and palate can be made to approximate those of the noncleft group. Even after extensive therapy, a characteristic change in the upper jaw shape remains detectable. These deviations are both dentoalveolar and skeletal (**51**). Adequate development of the apical base could not be achieved. Orthodontic treatment can compensate for good dental appearance in many patients by correcting size (**52**), however, fundamental morphological differences (shape) can only be partially treated. It is reasonable to conclude that adequate correction of the shape is only likely to be achieved through a secondary surgical correction after growth has finished. This affects at least about one-fifth of the patients (**53**) where camouflage orthodontic treatment is not possible.

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Table I: Lower arch measurements and statistics.							
	Cleft [n=27]			Noncleft [n=27]			
Distance	Mean [mm]	SD [mm]	Shapiro-Wilk (p)	Mean [mm]	SD [mm]	Shapiro-Wilk (p)	p
PAW	46.9	10.09	0.000 *	48.4	2.28	0.864	0.986
AAW	33.1	7.29	0.000 *	34.5	1.82	0.005 *	0.965
Dim 1	17.2	1.42	0.239	17.2	0.82	0.954	0.834
Dim 2	29.0	2.53	0.226	29.7	1.25	0.188	0.226
Dim 3	52.5	2.48	0.357	54.9	2,99	0.029 *	0.003 *
Dim 4	17.1	1.70	0.096	17.4	0.68	0.641	0.473
Dim 5	28.7	2.74	0.025 *	29.7	1.21	0.997	0.355
Dim 6	52.6	2.79	0.459	55.4	2.81	0.272	0.001 *
Dim 7	8.7	1.48	0.010 *	9.0	1.12	0.534	0.545
Dim 8	20.9	2.39	0.170	22.4	1.50	0.554	0.009 *
Dim 9	44.9	2.60	0.701	48.4	3.11	0.148	0.000 *
Dim 10	29.4	2.39	0.765	29.5	1.27	0.964	0.888
Dim 11	39.6	3.58	0.394	38.9	2.18	0.075	0.454
Dim 12	54.1	3.28	0.402	52.7	2.06	0.216	0.061

Table II: Upper arch measurements and statistics.							
	cleft			noncleft			
Distance	Mean [mm]	SD [mm]	Shapiro-Wilk (p)	Mean [mm]	SD [mm]	Shapiro-Wilk (p)	p
PAW	43.9	5.33	0.321	48.1	1.67	0.899	0.000 *
AAW	32.5	5.70	0.224	36.2	1.79	0.662	0.000 *
Dim 1	20.4	3.65	0.008 *	22.7	0.94	0.766	0.000 *
Dim 2	31.9	3.91	0.781	34.9	1.31	0.995	0.000 *
Dim 3	50.4	4.00	0.123	56.5	3.02	0.665	0.000 *
Dim 4	19.5	3.52	0.708	22.4	0.92	0.583	0.000 *
Dim 5	31.5	4.30	0.037 *	34.9	1.45	0.957	0.001 *
Dim 6	50.7	3.88	0.776	56.9	2.60	0.625	0.000 *
Dim 7	8.9	2.07	0.157	10.7	1.08	0.454	0.000 *
Dim 8	23.7	2.77	0.192	26.6	1.66	0.349	0.000 *
Dim 9	57.6	13.44	0.004 *	51.9	3.27	0.406	0.225
Dim 10	32.2	4.08	0.112	36.4	1.57	0.395	0.000 *
Dim 11	41.6	5.91	0.063	45.2	1.48	0.204	0.004 *
Dim 12	43.2	3.71	0.107	45.1	3.70	0.734	0.073

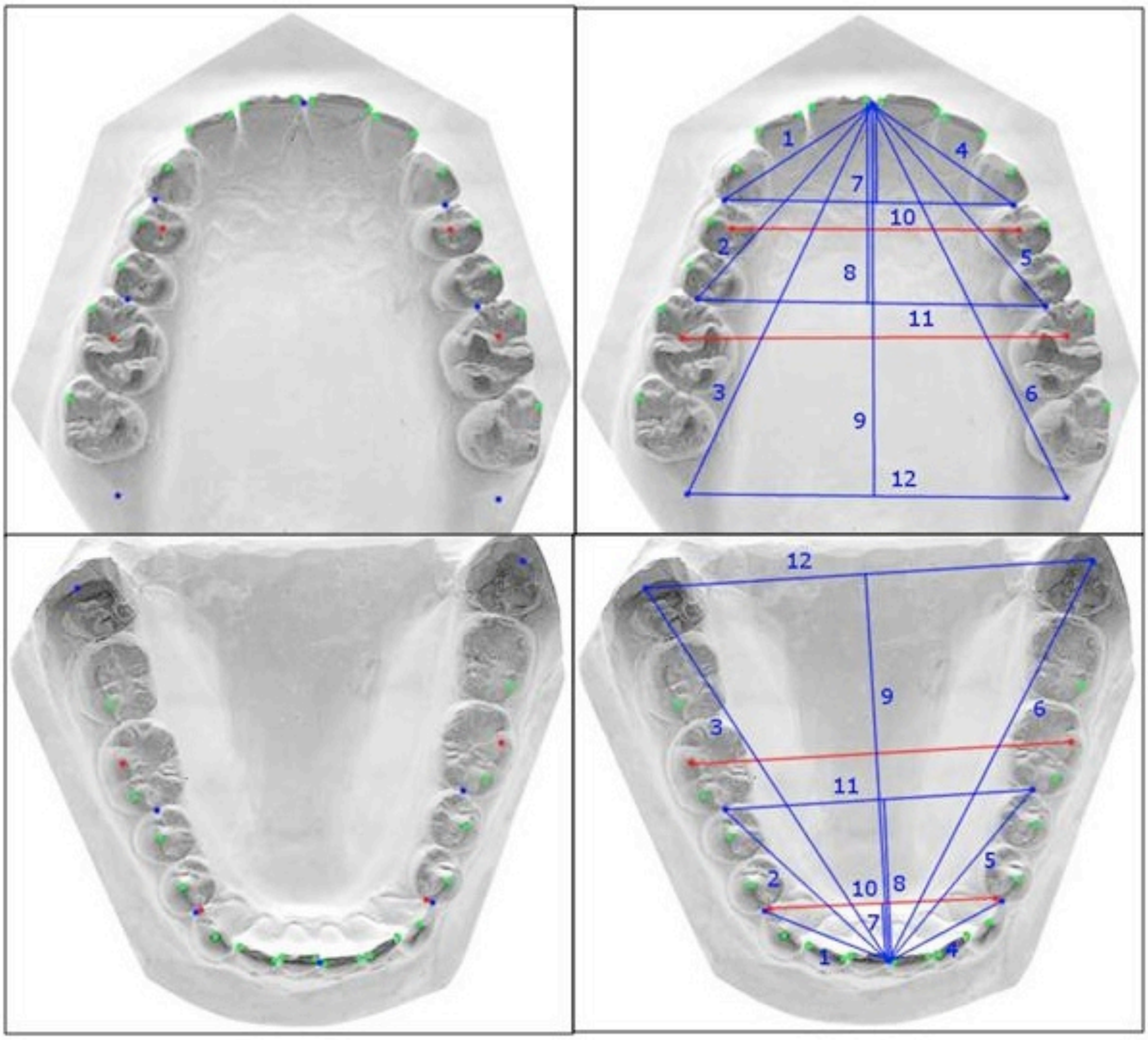

Figure 1: Landmarks and calculated distances for the upper and lower jaw (except the apical base landmarks). The dimensions Dim 1-12 according to Sillman (blue), Korkhaus measurements (red) and other morphological landmarks (green) are highlighted.

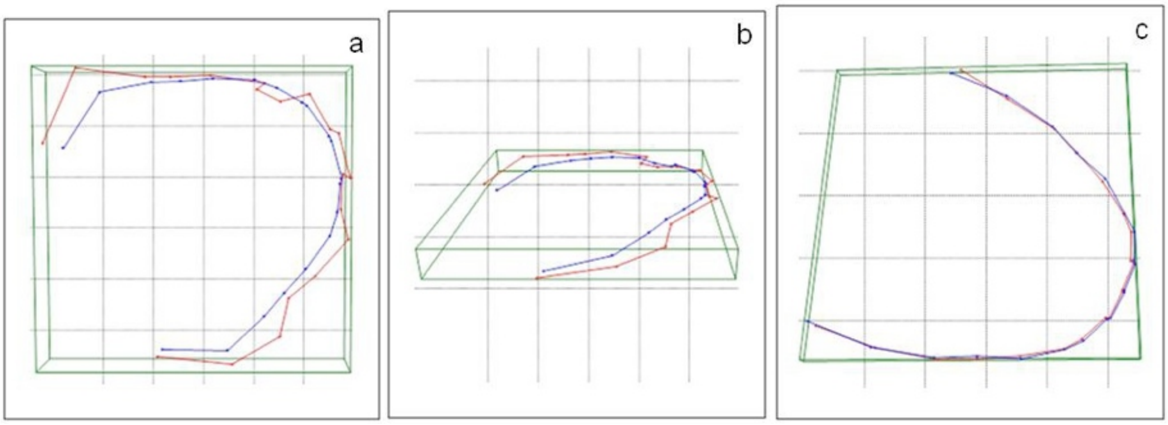

Figure 2: Superposition of the consensus shapes for the upper arch, top view (a), upper arch view from the right side (b) and mandible (c) after Procrustes transformation.

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