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Nasolabial shape and aesthetics in unilateral cleft lip and palate: an analysis of nasolabial shape using a mean 3D facial template

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Abstract. The aim of this study was to determine the amount of deviation in nasolabial shape in patients with a cleft compared with an average non-cleft face, and to assess whether this difference is related to nasolabial aesthetics. Three-dimensional stereophotogrammetric images of 60 patients with a unilateral cleft were used. To quantify shape differences, four average non-cleft faces were constructed from stereophotogrammetric images of 141 girls and 60 boys. Three-dimensional shape differences were calculated between superimposed cleft faces and the average non-cleft face for the same sex and age group. Nasolabial aesthetics were rated with the modified Asher-McDade Aesthetic Index using a visual analogue scale (VAS). Mean VAS scores ranged from 51.44 to 60.21 for clefts, with lower aesthetic ratings associated with increasing cleft severity. Shape differences were found between cleft faces and the average non-cleft face. No relationship was found for the VAS, age, and sex, except that a lower VAS was related to a higher nose and lip distance between the superimposed cleft and average non-cleft faces for nasal profile (P = 0.02), but the explained variance was low (R^2 = 0.066). In conclusion, except for nasal profile, nasolabial aesthetics were not influenced by the extent of shape differences from the average non-cleft face.

Key words: cleft lip; cleft palate; aesthetics; face; three-dimensional imaging; photogrammetry.

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The treatment of patients with cleft lip and palate (CLP) involves several surgeries aimed at anatomical, functional, and aesthetic correction of the congenital malformation. The ultimate treatment goal for these patients, however, must be to create a happy healthy individual whose deformity is not an issue in their personal life. Deviation from the average may affect the patient's wellbeing; therefore, clinicians try to improve the patient's appearance without cleft stigmata. Facial appearance plays a role in the social interactions between individuals and influences a person's perception of others[1,2]. It may also influence the quality of life (QoL) of the affected person[2–4].

Deviation from the average is difficult to measure. Two-dimensional analysis of the face, as performed in the past, is of limited value. In contrast, three-dimensional (3D) imaging techniques are more adequate. Studies of facial asymmetry in patients with CLP using 3D imaging have shown that asymmetry is most apparent in the centre part of the face, including the nose, lips, and partly the midface[5–7]. In a study using 3D analysis and stereophotogrammetry, it was shown that preadolescent children with different types of unilateral cleft had more facial asymmetry in the midface, whereas controls showed more asymmetry in the chin area. When the unilateral cleft defect also involved bony structures, asymmetry in the midface was more prominent[7].

Patients without a cleft also show a wide range of asymmetry, yet no specific area seems to influence the aesthetic rating of facial appearance[8,9]. In contrast, asymmetry in the nasolabial area in patients with a cleft has been associated with a lower aesthetic ratings[5],9. However, it is not clear how the aesthetics are related to deviations from a non-cleft reference face. A way to study shape differences between the faces of patients with and without clefts is to construct a mean face of non-cleft individuals and then compare it with faces of patients with clefts. Using this method, the shape differences in the midface can be quantified. This could be very useful when attempting to improve the appearance of patients with orofacial clefts and avoid cleft stigmata. This study was performed to investigate the null hypothesis that a larger deviation in nasal and labial shape is related to a worse aesthetic nasolabial score in CLP patients.

Subjects and methods

Patients

This study was conducted at the Cleft Palate Craniofacial Unit of Radboud University Medical Center, Nijmegen, The Netherlands. The study was performed in accordance with the Declaration of Helsinki with regard to research in human subjects. The use of anonymous data gathered during routine patient care is in accordance with Dutch laws on medical research. A written statement was obtained from the institutional review board stating that this study does not fall within the remit of the Medical Research Involving Human Subjects Act (WMO).

A total of 60 patients with a unilateral orofacial cleft, born between 1998 and 2004, were included in the study. The inclusion criteria were: (1) Caucasian, (2) non-syndromic complete unilateral cleft lip (UCL, $n = 10$), unilateral complete cleft lip and alveolus (UCLA, $n = 23$), or unilateral complete cleft lip, alveolus, and palate (UCLAP $n = 27$); (3) presence of 3D stereophotogrammetric images of the face at 9– 11 years of age; and (4) all treatments received at the same cleft centre, according to the treatment protocol used until 2008[10]. Lip closure (Millard technique) and primary nose correction (McComb technique) were done at the age of 6–8 months, and soft palate closure (according to a modified von Langenbeck procedure) at the age of 12–14 months. These procedures were performed by two surgeons with more than 10 years of experience in cleft surgery. Hard palate closure was done at the same time as the bone grafting procedure for closure of the alveolar cleft, if present. The timing of bone grafting was based on eruption of the canine; this is usually done at age 9–11 years of age. All stereophotogrammetric images were obtained prior to the bone grafting procedure and hard palate palatoplasty.

The patient sample was divided into two groups: ≤10.5 years old (range 8.6–10.5 years) and >10.5 years old (range 10.6–12.3 years). The younger group consisted of 23 boys and 18 girls (8 UCL, 15 UCLA, and 18 UCLAP). The older group consisted of 14 boys and 5 girls (2 UCL, 8 UCLA, and 9 UCLAP). For comparison, a compound face was created from the control children (see below). For the compound faces, patients (141 girls, 62 boys) were selected from the files of the Department of Orthodontics, Radboud University Medical Center, Nijmegen, The Netherlands and the Department of Orthodontics, University of Bern, Switzerland. Inclusion criteria for the controls were (1) Caucasian and (2) a maximum overjet of 6 mm. Exclusion criteria were (1) congenital malformation, (2) forced bite with lateral displacement of the mandible, (3) juvenile idiopathic arthritis, (4) impacted canines, and (5) a negative overjet.

3D image acquisition and processing

Three-dimensional facial images of patients and controls were taken with a 2-pod camera set-up for stereophotogrammetric imaging (3dMDface System; 3dMD LLC, Atlanta, GA, USA) with spatial resolution of 0.2 mm under standardized conditions. Images were taken in natural head position, and the patients were asked to keep their eyes open and to relax their facial musculature. All images were obtained by the same operator who has taken more than five such images a week for more than 5 years. All right-sided clefts were mirrored to ensure that, for calculations, all clefts were on the same side, i.e. the left side of the face. Confounding regions (neck, ears, and hair) in the images were removed using 3dMDpatient v3.1.0.3 software (3dMDpatient Software Platform; 3dMD LLC). The adjusted 3D photograph was imported into Maxilim software (Medicim NV, Mechelen, Belgium).

For the older age group (>10.5 years), the mean of 97 faces of girls and 41 faces of boys were used for the compound faces of girls and boys, respectively (age range 10.6–12.10 years). For the younger age group (≤10.5 years), the compound faces for girls and boys were created using the mean of 44 girl faces and 21 boy faces (age range 8.10–10.5 years). The compound face was created as follows: the images were pre-aligned using five landmarks (left and right endocanthion and exocanthion and subnasale); if necessary, additional manual positioning was performed. The polygon meshes of the 3D stereophotogrammetric images of all controls of the same sex and age group were superimposed to create the four compound faces using an iterative closest point (ICP)-based algorithm[11,12].

For measuring shape differences between the compound face and the cleft faces, the faces in the 3D images were separated into the nose and lip areas using the method described by Kuijpers et al.[7]. The chin and forehead were used by a surface registration algorithm (ICP)[12]. The areas of superimposition were defined using a grid (Fig. 1). A distance map was created between the compound face and the cleft face. For each patient, the mean and standard deviation (SD) for that distance map were used as the outcome. The measurements from the distance maps of the nose and lips were imported into MATLAB software version 7.4.0

(R2007a) (MathWorks Inc., Natick, MA, USA) to calculate the absolute mean distances and the 95th percentiles of the shape differences in the nasolabial area.

VAS scores

Nasolabial aesthetic scores for the patients with a cleft were assessed by eight orthodontic residents (four men and four women in their second year of training; age range 25–31 years) who had no experience with CLP treatment. The raters were instructed on how to manipulate the 3D image in all directions on the computer. They rated the nasolabial aesthetics of the 3D images (Fig. 2) using the modified Asher-McDade Aesthetic Index[13]. The index measures four nasolabial components: nasal form (NF), nasal deviation (ND), vermillion border (VB), and nasolabial profile (NP). Since the VB of the Asher-McDade Index cannot be validated as a distance, we did not use this component in the comparison. In addition, a 100-mm visual analogue scale (VAS) (score 0–100, from 'not aesthetically pleasing' to 'aesthetically pleasing') was used for all components[9], instead of the original five-point scale. The mean VAS score of the three nasolabial components (NF, ND, and NP) was used as the overall aesthetic score. Intra-rater and inter-rater reliability were assessed as described previously[9].

Statistical analysis

Absolute distance values between the superimposed surfaces of the cleft and compound faces were calculated for the nose, lips, and nose and lips combined. Descriptive statistics were used to define differences between the cleft and compound faces regarding the lip and nose on the cleft and non-cleft sides. Scatter plots were drawn for the VAS scores and absolute distances per cleft type to assess if the relationship between the two was dependent on the cleft type and if all cleft types could be combined in the regression analysis. A regression analysis was performed for the mean VAS for NF and absolute distance of the complete nose, the mean VAS for ND and absolute distance of the complete nose, the mean VAS for NP and absolute distance of the complete nose, and mean VAS score for the NF–ND–NP and absolute distance of the complete nose and lip. The distances were the dependent variable in all four analyses and the VAS score was used as an independent variable. As age and sex can be potential confounders, they were corrected for by adding them to the model as independent variables.

All statistical analyses were performed with IBM SPSS Statistics version 22 (IBM Corp., Armonk, NY, USA) by the same statistician (EB). The significance level was 0.05. For all models, the residuals were tested using the Kolmogorov–Smirnov test. In all cases, a lack of normality was not found.

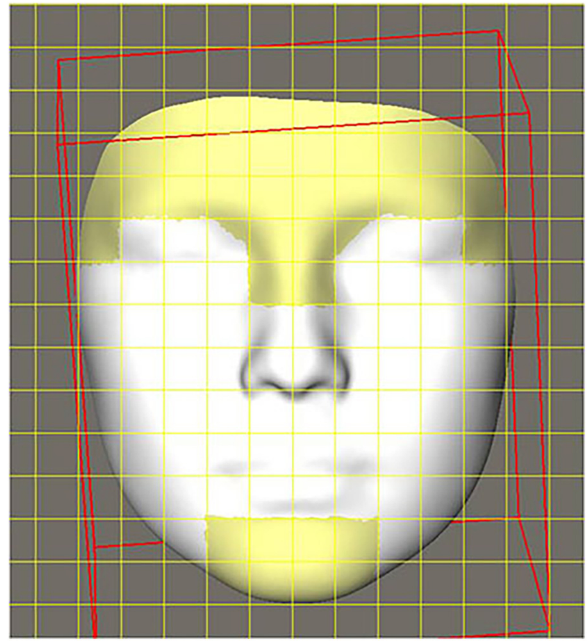

Fig. 1. Areas for superimposing each cleft face on the compound face. Yellow areas used for basic superimposition.

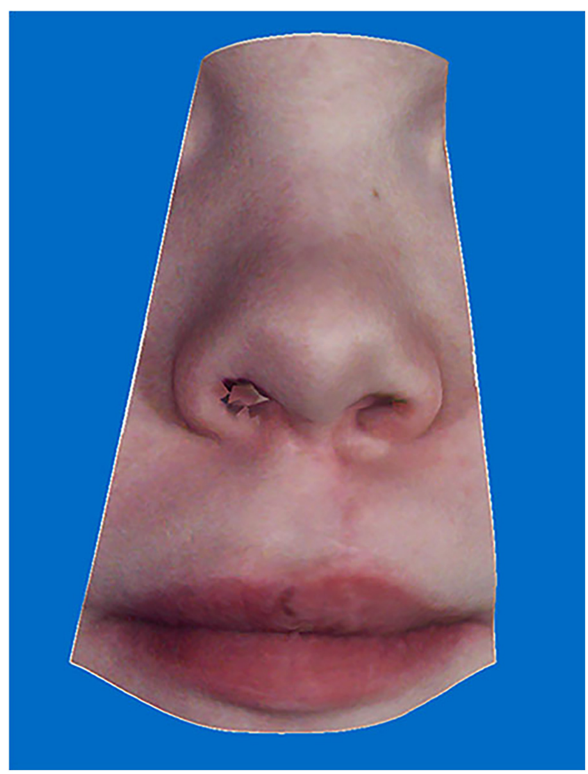

Fig. 2. Nasolabial 3D stereophotogrammetric image used for aesthetic rating. This image could be manipulated in all directions.

Results

The mean aesthetic scores for the three nasolabial components separately (NF, ND, and NP) and the overall aesthetic scores (NF–ND–NP) in subjects with UCL, UCLA, and UCLAP are presented in the Supplementary Material (Table S1). The mean overall aesthetic scores (mean VAS for NF–ND–NP) for the three cleft groups ranged from 51.44 (SD 8.79) to 60.21 (SD 8.41), with a lower aesthetic rating corresponding with increasing cleft severity. The same pattern was observed for the three aesthetic components of the nose separately.

The mean distances between the cleft and compound non-cleft faces for the three different cleft types and two age groups are presented in the Supplementary Material (Table S2). The largest shape differences, which were mostly >4.0 mm in both age groups, were found for the combined nasolabial area. The mean of the standard deviations was high (age ≤10.5 years: range 3.06–3.44 mm; age >10.5 years: range 2.53 to 3.60 mm).

Scatter plots were used to analyse whether the relationship between the 3D score (the absolute distance between the cleft and compound face) and the VAS score depended on the cleft type (Supplementary Material, Fig. S1). The scatter plots showed that the relationship between the two was not influenced by the cleft type; hence, the results of all cleft types were grouped together for further analysis. The results of the regression analysis are shown in Table 1. The overall aesthetic score, sex, and age had no influence on the absolute distance of the deviations of the nose and upper lip, and the VAS scores for NF and ND were not related to the absolute distance of the deviations of the nose. The only significant relationship was for the VAS for NP: a lower VAS score for NP was associated with a higher absolute distance for the deviation of the nose and lips; however the explained variance was very low ($R^2 = 0.066$).

Discussion

The goal of CLP treatment is to restore the anatomy and function of the oral facial region and create a facial appearance that is as normal as possible, with the least cleft stigmata. To determine the deviation from the average, cleft faces were compared with 3D compound faces of non-cleft individuals for boys and girls in two age groups. Multiple compound faces were used, because potential differences in facial morphology between sexes and ages has been demonstrated[14,15] and facial growth deformities in the cleft group may express differently over time. The rationale to divide the sample into 'younger' and 'older' groups was that differences in facial morphology before pubertal growth for boys and girls are minimal, but may occur in the older age groups. Therefore, a cut-off age of 10.5 years was chosen.

The method used in this study to create the compound faces, based on 3D stereophotogrammetric images, was described by Kau et al.[14,15] and has been used in several studies[16–18]. However, most of these studies were performed on adult patients. A systematic review comparing different 3D imaging techniques demonstrated that 3D stereophotogrammetry in younger children is a reliable and accurate method to quantify soft tissue-based facial dimensions[19]. However, non-cleft compound faces still have to be developed for a wide range of different age groups and ethnicities in order to assess, at the individual patient level, how the face of a patient with a cleft differs from a mean non-cleft face.

Table 1. Results of regression analyses using the mean VAS for different components of the nasolabial region, sex, and age as independent variables, and the absolute distance between the compound face of non-cleft individuals and the superimposed cleft face as the dependent variable.

Estimate of effects

Dependent variable	Independent variable	Effect	95% CI	*P*-value	Adjusted R^2
Absolute distance nose and lip	Constant	5.648	3.833 to 7.463	<0.001	
	Mean VAS NP, NF, ND	−0.018	−0.049 to 0.013	0.250	
	Sex	−0.401	−1.103 to 0.300	0.257	
	Age	0.154	−0.580 to 0.887	0.677	
					−0.005
Absolute distance nose and lip	Constant	6.289	4.809 to 7.770	<0.001	
	Mean VAS NP	−0.031	−0.057 to −0.005	0.020	
	Sex	−0.374	−1.051 to 0.303	0.273	
	Age	0.174	−0.533 to 0.881	0.624	
					0.066
Absolute distance nose	Constant	2.300	1.537 to 3.064	<0.001	
	VAS NF	−0.001	−0.014 to 0.012	0.893	
	Sex	−0.293	−0.667 to 0.052	0.093	
	Age	0.086	−0.290 to 0.046	0.650	
					0.000
Absolute distance nose	Constant	2.553	1.606 to 3.500	<0.001	
	VAS ND	−0.005	−0.021 to 0.010	0.507	
	Sex	−0.315	−0.674 to 0.044	0.084	
	Age	0.080	−0.294 to 0.455	0.669	
					0.007
Absolute distance nose	Constant	2.414	1.536 to 3.292	<0.001	
	Mean VAS NF and ND	−0.003	−0.017 to 0.012	0.698	
	Sex	−0.309	−0.669 to 0.050	0.090	
	Age	0.083	−0.293 to 0.459	0.659	
					0.002

VAS, visual analogue scale score; CI, confidence interval; NP, nasolabial profile; NF, nasal form; ND, nasal deviation.

The compound faces were used to address the research question of whether a larger deviation in nasal and labial shape is associated with a worse aesthetic score for the nasolabial region in patients with CLP. A previous study showed an increase in VAS scores, meaning better aesthetics, when the symmetry of the nasolabial area increased[9]. However, in that study, a quantitative assessment of the asymmetry in the nasolabial area was only done for cleft faces and not for non-cleft faces. In a follow-up study using 3D stereophotogrammetry, we found that preadolescent children with unilateral clefts showed significantly more facial asymmetry than children without clefts, especially in the nasolabial area. The nose was the most asymmetric region in UCLA and UCLAP patients[7]. In the present study, the shape differences of the nasolabial area were quantified by comparing cleft faces with a mean compound face of boys and girls of two age groups to investigate whether the shape difference would affect aesthetic ratings. Superimposing the cleft faces on the compound faces was done by surface registration. As most differences were expected in the nasolabial area, superimposition was done on regions outside this area. Surface registration is facilitated when using curvatures in a face as far as possible from each other; therefore, the eyebrow region was used together with the chin.

Shape differences in the nose and lips compared with non-cleft controls were found. These differences, however, did not affect the VAS aesthetic rating, except for the nasal profile. In this study, the vermilion border scoring used in the Asher-McDade Aesthetic index was not included, because the vermilion border cannot be measured as a shape difference. This may have changed the mean VAS slightly. However, nasal shape seems to have the greatest impact on satisfaction with the result[5,20,21]; therefore, excluding the vermillion border may be of minor importance. The Asher-McDade Aesthetic Index was used because it is considered superior to other systems in non-syndromic CLP[22]. Instead of the five-point scale used in the original Asher-McDade Aesthetic Index, it was decided to use a VAS, as it was felt that this was more reliable and sensitive than a categorical scale[23,24]. Ratings were done by postgraduates who had just started their training in orthodontics. Even though they had not been involved in cleft palate treatment, their dental background may have had an influence on the VAS. However, a recent systematic review concluded that it remains unclear whether professionals and laypersons rate the facial appearance of patients with clefts differently. Some studies have shown that professionals are more critical[25], whereas others have shown the opposite[26]. Thus, the background of our layperson group probably did not affect the VAS results.

No correlation was found between the aesthetic rating and the magnitude of the shape difference between patients with clefts and controls. In 2016, Kaipainen et al.[8] assessed the amount of facial asymmetry in non-cleft individuals and tried to clarify the relationship with facial attractiveness. They found comparable results to those of the present study, i.e. they were not able to show an effect of asymmetry on facial appearance. This is in contrast to Meyer-Marcotty et al.[27], who found lower aesthetic scores when asymmetry was present in the midfacial area, while asymmetry in the outer regions of the face had less influence on aesthetic scores. This may have been influenced by the scoring method. In the study of Meyer-Marcotty et al.[27], the whole face was visible, whereas in the present study, aesthetic ratings were performed on the nasolabial area without showing the rest of the face. As our focus was on nasolabial aesthetics, other facial features were eliminated from the 3D stereophotogrammetric images. In all previously mentioned studies, aesthetics were measured on static records. However, facial aesthetics can be different at rest and during function. Four-dimensional stereophotogrammetry could provide further insights, because functional facial differences between patients with clefts and controls may become apparent with this method[28]. Further studies must be performed to understand the complex relationship between facial morphology and aesthetics.

There are some limitations to this study. The number of patients with a cleft lip only was small and differences from the average face should be interpreted with caution. Both the patients and the raters in this study were Caucasian. We were not able to create a compound non-cleft face for other ethnic groups due to a lack of data. It was decided to exclude all other ethnicities from the patient group, as studies using 3D soft tissue imaging have shown differences in facial morphology between ethnic groups[29]. The compound faces were created using the ICP algorithm[30]. This was to minimize outlying points[12]. It was decided to divide the control group into two different age groups because we expected differences in facial morphology between ages. This resulted in a smaller sample per age from which the average faces were developed, which may have influenced the range of shape differences. Our focus was on nasolabial aesthetics; therefore other facial features were excluded from the scoring, but this may have influenced the raters' perceptions[23]. Furthermore, this was a scoring of aesthetics on static 3D images. Aesthetics may be perceived differently in four dimensions, for example during smiling and speaking[28].

In conclusion, under the conditions of this study, it is concluded that the perception of nasolabial aesthetics is not influenced by the extent of shape differences from the average non-cleft face, except for nasal profile. Thus, factors other than nasolabial deviation may influence the rating of nasolabial aesthetics. Future research should focus on identifying these potential factors as they may contribute to higher treatment satisfaction for our patients.

Competing interests

None.

Funding

No funding was received.

Ethical approval

The study was performed in accordance with the Declaration of Helsinki with regard to research in human subjects. The use of anonymous data gathered during routine patient care is in accordance with Dutch laws on medical research. A written statement was obtained from the institutional review board stating that this study does not fall within the remit of the Medical Research Involving Human Subjects Act (WMO).

Patient consent

For this type of study, formal consent was not required.

Statement to confirm

This is to confirm that all authors have viewed and agreed to the submission.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.ijom.2020.06.003.

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