3D comparison of average faces in subjects with oral clefts

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SUMMARY This prospective cross-sectional, case-controlled morphometric study assessed three dimensional (3D) facial morphological differences between average faces of 103 children aged 8–12 years; 40 with unilateral cleft lip and palate (UCLP), 23 with unilateral cleft lip and alveolus (UCLA), 19 with bilateral cleft lip and palate (BCLP), 21 with isolated cleft palate (ICP), and 80 gender and age-matched controls. 3D stereophotogrammetric facial scans were recorded for each participant at rest. Thirty-nine landmarks were digitized for each scan, and x-, y-, z-coordinates for each landmark were extracted. A 3D photorealistic average face was constructed for each participating group and subjective and objective comparisons were carried out between each cleft and control average faces. Marked differences were observed between all groups. The most severely affected were groups where the lip and palate were affected and repaired (UCLP and UCLA). The group with mid sagittal palatal deformity and repair (ICP) was the most similar to the control group. The results revealed that 3D shape analysis allows morphometric discrimination between subjects with craniofacial anomalies and the control group, and underlines the potential value of statistical shape analysis in assessing the outcomes of cleft lip and palate surgery, and orthodontic treatment.

Introduction

Cleft lip, isolated cleft palate, and cleft lip and palate are among the most commonly occurring congenital craniofacial anomalies (Sampson et al., 1997), with approximately 1 in 600 new born babies affected worldwide (World Health Organization, 2002). In the UK, the incidence on average is 700 new cases of orofacial clefting each year, accounting for 65 per cent of all congenital craniofacial deformities (Williams et al., 1994), and this is higher in the northern region of England at 1 per 440 live births as reported by the Office of National Statistics (HMSO) in 2005.

Cleft repairs are usually performed early in life. The main aim of surgical repair is the restoration of normal facial aesthetic and function (Hodgkinson et al., 2005). Recent development of three-dimensional (3D) assessment tools and the innovation of statistical shape analysis offer new improved methods to measure severity and outcome.

Accurate discrimination between individuals with craniofacial anomalies and controls can be achieved by comparing facial morphology of average faces of each group (Hammond et al., 2004). This allows assessment of subjects with oral clefts, to matched control groups from the same ethnicity, to plan and refine surgical repair. Construction of average faces from laser scanned images has been reported in the literature. Toma et al. (2008) constructed an average face for 15-year-old females and males, Kau et al. (2006) used average faces as a template for the comparison of facial skeletal disproportions, and Gor et al. (2010) and Božić et al. (2009) used this approach to discriminate between male and female average faces of two different Caucasian populations.

Duffy et al. (2000) reported preliminary work where average faces were constructed from 3D models of unilateral cleft lip and palate (UCLP; 10), unilateral cleft lip and alveolus (UCLA; 9), bilateral cleft lip and palate (BCLP; 7), isolated cleft palate (ICP; 13), and matched 25 controls. Each average face with a cleft was superimposed on the control, and a colour-coded image employed to subjectively compare the superimposed faces. In their method of constructing average faces, the total image acquisition time was 7.5–10 seconds, which could introduce errors as facial expression can be altered. Furthermore, each scan was generated by aligning separate right and left scans. Duffy et al. (2000) concluded that the superimposition technique offered a superior quantification for the evaluation of treatment outcome. Recently, Bugaighis et al. (2011) compared constructed photorealistic average faces of 39 male and 41 female, 8–12-years-old subjects, without clefts, using the same method described in this study.
The aim of this study was to compare 3D facial forms of children with UCLP, BCLP, UCLA, ICP, and matched control group, using 3D data acquisition and advanced assessment tools.

Materials and methods

Ethical approval was granted by the Northumberland Local Research Ethics Committee (06/Q0902/36) and informed consent was obtained from parents/guardians and children. This investigation was conducted on 103 children aged 8–12 years with non-syndromic operated: 40 with UCLP (mean age: 10.1 years), 23 with UCLA (mean age: 10 years), 19 with BCLP (mean age: 10 years), ICP 21 (mean age: 10 years), and 80 gender and age-matched controls (mean age: 10.5 years). All patients were from North East England and Caucasian in origin. Children with non-syndromic oral clefts were recruited from the cleft multidisciplinary clinics at the Royal Victoria Infirmary co-ordinated clinics in the North East of England. All clefts in children with UCLP were complete. The clefts were repaired prior to the implementation of the Clinical Standards Advisory Group (CSAG, 1998) recommendations where five different surgeons using different protocols were involved in cleft repair during the first year of the children’s lives. The exact surgical procedures followed could not be identified due to poor documentation at that time. None of the participants had started significant orthodontic treatment. Any child who had performed alveolar bone grafting was excluded.

A control group was recruited from the Child Dental Health Department at Newcastle Dental Hospital; all controls had harmonious balanced faces, class I dental occlusion, competent lips, and no craniofacial abnormalities, including hypodontia, and none had undergone orthodontic treatment. Any child who had performed alveolar bone grafting was excluded. A control group was recruited from the Child Dental Health Department at Newcastle Dental Hospital; all controls had harmonious balanced faces, class I dental occlusion, competent lips, and no craniofacial abnormalities, including hypodontia, and none had undergone orthodontic treatment. The control group was sufficiently large to have a 95 per cent power to detect an effect size (standardized difference) of 0.85, assuming a type I error rate of 5 per cent (Field, 2005). Separate average faces of males and females could not be conducted because of the small number of subjects in all cleft groups. The same sample was employed to assess shape variation within, and between, the studied groups using geometric morphometrics (Bugaighis et al., 2010).

Image acquisition and processing

Scans were captured at rest using a 3D stereophotogrammetry system (3dMD, Atlanta, USA). Children were positioned at 95 cm in front of the unit, with the Frankfort plane raised anteriorly by 10 degrees to the horizontal, to ensure a clearer picture of the nose. Subjects were instructed to be at rest, with lips lightly opposed, if possible without straining, and eyes to be open without stretching the forehead. The acquisition time was 2ms that minimized changes in position or facial expression. All cameras and flashes were synchronized to capture the entire surface of the face from ear to ear simultaneously, generating one continuous point cloud. Four geometrical and two texture images were recorded for each child. Subsequently, the captured images were processed to generate a triangulated point cloud, constructed from the captured images in less than a minute. The resulting 3D image comprises approximately 40,000 vertices for a typical captured face, mapped with two megapixel texture image.

Landmark identification

MorphAnalyser software (V 2.07, user.aber.ac.uk/bpt) is a software package originally developed by Tiddeman et al. (2000) for the 3D visualization of faces, interactive placement of correspondent landmarks, and extraction of the x-, y-, z-coordinates of each landmark. Thirty-nine anthropometric landmarks mainly based on those of Farkas (1994) were recorded on each image to characterize facial, and especially nasolabial form (Supplementary Table 1 and Supplementary Figure 1). From these, average faces were constructed by matching corresponding surfaces between specified landmarks.

Orientation of images imported to the software was achieved by building a Cartesian coordinate system. All scans with right unilateral clefts were reflected using MorphAnalyser software, in order to keep the cleft side always on the left for analysis consistency. The face was standardized by creating a plane using the three first recorded landmarks (Bookstein, 1987). For this study, sublabialis landmark (sl), was chosen as origin, with exocanthion right (exR) and exocanthion left (exL) as second and third landmarks for standardization. These landmarks were selected to span the area of interest (nasolabial region). Subsequently, the oriented image was saved as .obj format and a joint.jpeg file. The x-, y-, z-coordinates of each landmark were saved as a template in a text file. Hence, for every scan a folder including the three file formats was created for further use.

The software was validated by an error assessment study. The overall error from image acquisition, MorphAnalyser software, and operator error was on average 0.5 mm. This value was comparable with other 3D morphometric validation studies (Garrahy, 2002; Hajeer, 2003).

Construction of average faces

Construction of an average face for each cleft or control group separately was achieved by:

- Importing a highly accurate 3D image with its template by MorphAnalyser as a base mesh for each average face.
- Optimal superimposition of the images, by translating, rotating, and scaling the images one at a time so they best fit and to minimize residual spaces between the correspondent landmark configurations (generalized Procrustes analysis). Hence, the landmarks were used to guide the alignment between the correspondent images.
but the dense alignment occurred between all the vertices of the image so they serve as landmarks. This aids in the precision and accuracy of the construction of the average face and explains the display of parts of the image in which landmarks were not digitized such as the cheeks.

Finally, the 3D average face of the data set was displayed on the screen, and the corresponding files were saved in two formats: .avg and .obj. The overall folders preparation and processing time for a group of 21 subjects (ICP group) took half an hour. Shape variation was investigated by the superimposition of each of the average faces of the cleft group on the control average face using surface-based registration with 3dMD software. A colour-coded image displays the subjective differences between the two surfaces. Red shows the most positive distance of the superimposed surface (average face from cleft groups) compared with the base surface (control). Green shows the most negative, and blue indicates well-aligned surfaces. A histogram presents the descriptive statistical differences in millimetres (Figure 2). The difference was examined subjectively using the colour-coded image.

An error study was conducted to validate averaging and superimposition methodology. Ten subject’s images from each group were submitted to averaging to construct five average faces, one for each group. The same procedure was repeated again after 1 week with the second set of images, and average faces were constructed for a second time. Each average face was superimposed on its correspondent, to assess objectively and subjectively the differences between the average faces. The superimposition showed a homogeneous facial surface colour, indicating identical average models. Objective differences were displayed in the histogram; the average mean value, standard deviation (SD), and the root mean square (RMS) distances were equal in both trials.

### Results

**Difference in average face of control compared with UCLP**

The UCLP average face had a wider, more asymmetric nose, with a flattened dome of the nasal crura, which is deviated towards the unaffected side (Figure 1). The mouth was asymmetric, and the cheeks and zygomatic regions were flatter. The UCLP average face presented with a straighter profile compared with the control face.

The frontal view of the colour-coded image (Figure 2) shows the nasal left and right root surfaces in green that describes as retruded, though wider base of the nose in the affected side is shown in green colour even though the base of the nose is retruded in the UCLP average face (green colour). The surface of the nasal tip of the control average face was red, indicating that this area is more protrusive in the UCLP group. This is the area where the nasal tip of the control superimposes on the left alar wing of the UCLP average face, due to the deformed nasal dome and its displacement towards the unaffected side. Although, it is generally accepted that the tip of the nose appears flatter and more retrusive in clefts where the lip is involved. A possible explanation is that the average nose might...
be larger in the UCLP group compared to control. The philtrum surface is protruded in the UCLP average face (red colour). This is the place of the shallow philtrum furrow in the control average face, which is flattened in the UCLP average face. The cheek surface was asymmetric, with the left (affected) UCLP cheek surface being more retruded than the right side. The mandibular surface is retrusive in the UCLP average face extending laterally, where the left side is more retrusive than the right side. It can be seen in the left profile view, that the right unaffected nasal region, and right nasal rim surface, was retrusive in the UCLP face (green colour). The surfaces of the left nasal side of both average faces were better correlated than the right side. The UCLP average face had a reduced posterior face height and retruded left facial surface (green colour). There was also a difference on the right side, but to a lesser extent.

The histogram (Figure 2) shows that for the 8536 valid distance measurements displayed, differences between both facial surfaces are more on the green coloured side, emphasizing the generally retrusive nature of the UCLP average face. The RMS value of the distance measurements was 2.8 mm, with similar variability to BCLP (SD = 2.2 mm), but greater than ICP (SD = 1.3 mm) and UCLA (SD = 1.7 mm) average faces compared with the control.

**Figure 2** A colour-coded image (A) of the UCLP average face (difference) superimposed on control average face (reference) and a histogram (B) showing differences between both images.

**Difference in average face of control compared with UCLA**

Figure 3 shows that differences between the control and UCLA average faces were asymmetric, and mainly found in the nasolabial region. Although, these differences were less than those between the control and UCLP average faces. The UCLA average face had a more asymmetric nose and upper lip. The dome of nasal crura was deviated towards the unaffected side as well as being slightly flattened at the nasal tip. The profile view shows a slightly flattened upper lip and mid-facial region, although more similar to control than the differences between the control and UCLP profile.

The UCLA average face superimposed on the control (Supplementary Figure 2) shows that most regions, excluding the nose and upper lip, were similar (bluish in colour). The nasal root surface and the insertion of the alar wings in the upper lip were retrusive in the UCLA average face (green colour). The affected left nasal surface, nasal bridge, and the surface of the nasal dome were mostly protrusive in the UCLA average face. The philtrum surface was protruded (red colour) close to the lip repair surface.

The profile views show minor asymmetry between both right and left superimposed average surfaces. The right side approximated well between both the UCLA and the
control but they did not coincide, whereas the left facial surfaces in both average faces correlated well (blue colour).

The histogram curve is mostly in the blue coloured area confirming that both faces were similar. However, some data elements were in the red and green coloured areas due to the retrusive and protrusive relationships between the two surfaces as described above. There were 8536 valid distance measurements with a RMS value of 1.97 mm (SD = 1.72 mm).

**Difference in average face of control compared with BCLP**

The BCLP average face had a broader nose, more inferiorly positioned nasal dome and flatter zygomatic area and cheeks (Figure 4). The upper lip seemed to be flatter and retrusive. The profile views show a retrusive mid-facial region suggesting a class III BCLP profile. Superimposition of both faces highlights a retrusive nasal dome and upper lip in the BCLP average face (Supplementary Figure 3). On the other hand, both sides of the nasal bridge, extending to the soft tissue alar rims, were more protrusive in the BCLP average face (red colour). This feature was asymmetric with the left side being more protrusive. The surface between the mentolabial fold and the chin was more retrusive in the BCLP face (red colour). The right and left facial sides of both average faces were well correlated but did not coincide, and the asymmetry was less clear.

The histogram illustrates that for this colour-coded image, 8536 valid distance measurements were made with a RMS of 2.2 mm (SD = 2.2 mm). The curve was mainly in the blue coloured area indicating similar superimposed surfaces; that part of the curve in the green area relates to the retrusive surfaces in the UCLA average face.

**Difference in average face of control compared with ICP**

There were no apparent differences in the frontal view of the ICP and the control average faces (Figure 5). The profile view, however, reveals that the mandible was more retrusive, and the profile was slightly more convex, in the ICP average face.

Superimposition of both average faces (Supplementary Figure 4) shows that most of the nasolabial region, lower lip, mentolabial fold and chin were more retrusive in the ICP average face (green colour). On the other hand, the blue colour highlighted areas where the surfaces of both average faces were well correlated. The histogram illustrates that 9695 valid distance measurements were made with RMS of 1.5 mm (SD = 1.3 mm). Most of the curve was in the blue coloured area indicating similarity between the two superimposed surfaces, whereas that in the green area represents the retrusive surfaces in the ICP average face.

**Discussion**

This is the first study to apply advanced statistical tools to construct 3D photorealistic average faces, and to evaluate 3D facial form differences within and between the main cleft
groups. This allows greater understanding of the aetiology and influence of each cleft type on facial growth and development.

3D stereophotogrammetry is reliable and appropriate for the clinical assessment of facial morphometry in all three planes of space. Fast capture time, non-invasiveness nature, and ease of use make it child friendly. The display of a photorealistic 3D model of the face increases the accuracy of landmark digitization.

Construction of average faces offers an interesting perspective for diagnosis, treatment planning, and comparison
of facial shapes between different groups. This provides surgeons with valuable 3D templates that aids in planning the reconstruction of craniofacial anomalies. Orthodontists can use a soft tissue reference to understand soft tissue movement in relation to dentoalveolar and skeletal changes occurring with treatment and growth. This might develop to allow the prediction of facial growth for each gender and age group, instead of relying on 2D radiographic templates.

Superimposing 3D faces provides a relevant tool in the evaluation of orthodontic and surgical procedures by comparison of pre- and post-treatment facial images. Additionally, it can be employed in studying longitudinal soft tissue growth in cohort studies. The examined group’s average faces, where the alveolus is involved, can facilitate longitudinal studies of the consequences of growth and surgery such as the assessment of alveolar bone graft and orthognathic surgical techniques, on the facial shape of subjects with oral clefts. Furthermore, comparison of individual image of subjects with oral clefts with a control average face from the same ethnicity, gender, and age group, to aid in treatment planning.

The averaging and superimposition method is thus a promising assessment tool, which can be used as a practical diagnostic aid in the clinical setting, due to a fast, relatively simple, and non-invasive nature, this method requires greater development and refining. For example, the objective assessment technique employed in this study measures the closest point distances, between the two average shapes in millimetres, and offers an overall view of the two compared surface differences. However, those linear distance values cannot be used to precisely quantify those differences (Tiddeman et al., 1999).

The sample age group (8–12 years old) allowed was selected to examination of the effects of primary and secondary lip and palate surgical repairs. The age range was selected so that the subjects had not yet started significant orthodontic treatment at this stage, while at the same time, adequate growth and development had taken place to allow investigation of the treatment protocols undertaken. The age group was extended to five birth years (from 8 to 12 years) to overcome the problems arising of smaller sample sizes associated with studies of oral clefts (Mars, 2004). However, the sample size in the cleft groups was not sufficient to construct a separate male and female average face in each group.

Superimposition of the UCLP and UCLA average faces on the controls showed that differences were more pronounced between the UCLP and control than between the control and UCLA. The differences were asymmetric and were mainly in the nasolabial region. The philtrum furrow was more flattened and affected by the deformity in the UCLP than in the UCLA. It is generally accepted that the UCLP philtrum configuration is altered in the cleft cases, including the lip, and especially in the UCLP group. This is expected especially in cases operated before the implementation of the CSAG recommendations (CSAG, 1998), where a more heterogeneous methodology was undertaken.

The posterior face height and left facial surface were retruded in the UCLP face. It would be unrealistic to interpret these differences as only due to the asymmetry between both facial surfaces in the UCLP group, as it might have occurred due to altering facial expressions, while capturing the image for the individuals in both groups. Both right and left facial sides correlated well between the UCLA and the controls, although the right cheek and zygomatic surface was more protruded in the UCLA face. Duffy et al. (2000) reported retruded left cheek area in the UCLP average face, but it was not clear if this extended laterally to the left facial surface. Only frontal view images were shown in their study. They reported retruded nasal tip, and retruded affected clefted nostril, in the UCLP group. However, there was similarity between the controls and UCLA average faces in the nasolabal region, except for the left nasal surface. In this study, the surface of the control nasal tip superimposed on the deformed left alar surface was more protruded in both the UCLP and UCLA groups. This might have occurred due to the larger average nose in the cleft groups compared with the control. The protruded left nasal rim surface extended to the whole left nasal bridge surface in the UCLA face. The left alar surface was mostly similar in the UCLP and protruded in the UCLA average face. Further studies on different samples are required to confirm these observations.

The straighter profile in the BCLP group reported here was clearly seen by comparing the average faces of both the control and the BCLP average face. This was mostly due to a retrusive maxilla, confirmed by the retrusive upper lip surface, when both the controls and the BCLP faces were superimposed. The flattened nasal dome in the BCLP group was retrusive and the nose wider in the BCLP face.

With the absence of lip involvement, the ICP average face was the most similar to the control group. The superimposition of the ICP average face on the controls showed differences between both surfaces in the bimaxillary region, and especially the mandible, where the control soft tissue was more prominent in that region. A number of studies reported that ICP might occur as a consequence of a small retruded mandible, which might interfere with displacing the tongue inferiorly, to allow the palatal shelves to fuse during the eighth week of intra-uterine life (Kreiborg and Herman, 2002; Watson, 2004; Berkowitz, 2006; Kreiborg et al., 2006). Similar findings were stated in a cross-sectional radiographic study on unoperated Danish infants with clefts (Dahl et al., 1982), which found that infants with ICP had a more retrusive mandible compared with other cleft groups. The maxilla was retrognathic as well, but to a lesser extent than the mandible.

The findings of this study highlight the greater effect of lip and palate cleft defects, and post-repair sequelae, on facial shape than cleft palate alone (Dahl, 1970; Molsted et al., 1995). Moreover, these results underline the potential
value of statistical shape analysis in assessing the outcomes of cleft lip and palate treatment.

Conclusion

3D shape analysis allows comparison of morphometric differences between subjects with craniofacial anomalies and a control group, and underlines the potential value of statistical shape analysis, in assessing the outcomes of cleft lip and palate surgery and orthodontic treatment.

Supplementary material

Supplementary material is available at European Journal of Orthodontics online.

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