Three-dimensional quantitative evaluation of facial morphology in adults with unilateral cleft lip and palate, and patients without clefts

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Abstract

The aims of this study were to assess the quantitative values of measurements using proportion indices in the craniofacial region in patients with repaired, non-syndromic, complete unilateral cleft lip and palate (UCLP), and compare them with a control group who did not have clefts using the non-invasive systems of 3-dimensional technology. Three-dimensional measurements of the facial surfaces of 15 Malay patients who had UCLP repaired and 100 Malay control patients aged 18–25 years were analysed. The 3-dimensional images of the respondents’ faces were captured using the VECTRA-3D Stereophotogrammetry System. Eleven craniofacial proportions were assessed using a combination of 18 linear measurements obtained from 21 anthropometric soft tissue landmarks. These measurements were used to produce proportion indices to find the differences in the morphological features between the groups, and assessed using the independent sample t test and z scores. There were significant differences between the groups in 7 out of 11 craniofacial proportion indices ($p = 0.001–0.044$). Z scores of 2 indices were disproportionate. They were nasal index (which was severely supernormal) and upper lip index (which was moderately supernormal). Patients with UCLP had higher mean z scores, indicating that patients with UCLP tended to have larger faces than the control group. There were clinically important differences mainly in the nasolabial area, where the nose and the upper lip were wider, larger, or flatter in patients with UCLP.

Keywords: Cleft lip and palate; Facial morphology; Quantitative evaluation; Stereophotogrammetry; Three-dimensional analysis

Introduction

Quantitative assessment of patients with CLP that involves facial measurements between specified facial landmarks can be either direct on live subjects, or indirect using soft tissue radiography, plaster casts, 2-dimensional photographs, and 3-dimensional imaging. Craniofacial anthropometry is important in clinical assessments such as planning and evaluation of treatment and assessment of outcome in various medical fields, particularly in dentistry. Apart from that it is also useful in the diagnosis of syndromes, to estimate normal and abnormal growth, and plan and evaluate surgical or orthodontic treatment.

Traditionally, facial morphology has always been analysed through observation and visual examination to identify the craniofacial complexity in facial anomalies. However, direct measurement of subjects has many disadvantages. It...
is time-consuming, requires the cooperation of the patient, poses risks of injury, and usually identifies only the most obvious proportions on the face to find imbalance. Visual judgement is also influenced by the most striking disproportions but usually cannot isolate the factors that cause them.\textsuperscript{12} The calculation of indices and proportions were, therefore, introduced to estimate the quality of facial morphology. One of the uses of anthropometric proportion indices is in the measurement of facial attractiveness as an evaluation of surgical-orthodontic treatment.\textsuperscript{13}

Non-invasive 3-dimensional technology systems have been developed in dentistry over the last decade. Currently, 3-dimensional stereophotogrammetry imaging systems are more reliable and have become the gold standard.\textsuperscript{14} They provide surgeons, orthodontists, and others who deal with craniofacial structure with the opportunity to develop new approaches to the diagnosis and management of patients.\textsuperscript{15} Although there are many publications about anthropometric measurement of the craniofacial complex, there are not many that document the measurement on subjects with CLP using proportion indices and 3-dimensional technology.

The aim of this study therefore was to assess the quantitative values of measurements using proportion indices in the several craniofacial regions in patients with repaired, non-syndromic, complete unilateral cleft lip and palate (UCLP), and compare their results with those of a control group without clefts using non-invasive 3-dimensional technology systems.

Materials and methods

Sample

The study sample comprised 15 patients with UCLP (5 men and 10 women) aged between 18 and 25 years who had attended the Combined Cleft Lip and Palate Clinic at the Faculty of Dentistry, University of Malaya. Inclusion criteria were: age between 18 and 25 years and they had repaired, non-syndromic, complete UCLP. The lip had been repaired at 3 months of age (Millard and Delaire) and they had palatal surgery at 9 months of age (Von Lagenbeck and Two Flap), the operations done by either a plastic or an oral maxillofacial surgeon. In addition, they had no orthognathic surgery before imaging, and they were of second generation Malay ethnicity.

One hundred respondents who had not had clefts (50 men and 50 women) aged between 18 and 25 years were selected from one of the private colleges in Kuala Lumpur. Inclusion criteria were: age between 18 and 25 years; they had no facial clefts or other facial abnormalities; they had no previous orthodontic treatment; and they were of second generation Malay ethnicity.

Ethical and written approval for this study was obtained from the Medical Ethics Committee, Faculty of Dentistry, University of Malaya [DF CD1211/0059(L)]. Verbal and written consent was also obtained from the participants. A questionnaire was used to confirm that all parents and grandparents were Malays and there had been no inter-racial marriages.

Capture, measurement, and analysis of 3-dimensional images

All 3-dimensional facial images were captured using the VECTRA-3D dual module system for full-face imaging (Canfield Scientific Inc., Fairfield, NJ, USA). The cameras were calibrated before the image was captured using the manufacturer’s guidelines to ensure consistency and magnification. The subjects were seated 95 cm in front of the camera on a self-adjustable stool and each subject’s head position was seen by live video preview, which is one of the features of the VECTRA-3D. The 3-dimensional images of the subjects were produced with a capture time of two milliseconds/image, and the scanned images were then displayed and analysed with the 3D Mirror Software.

A total of 18 linear measurements derived from 21 anthropometric soft tissue landmarks were recorded (mm) from 4 regions of the craniofacial complex (face, orbits, nose, and orolabial area) (Fig. 1).\textsuperscript{12,16,17} Eleven craniofacial proportion indices were derived from the combination of these 18 linear measurements (Table 1).\textsuperscript{12,16} All measurements and analysis were made by one assessor.

Method error

Thirty 3-dimensional facial images were selected at random from each group, and landmarks were identified and measurements made after a 2-week interval. The measurements obtained were compared with the first measurements to assess the reliability of measurements from repeat identification of landmarks. The intraclass correlation coefficient test (ICC) was used to assess the reproducibility of the 2 readings. ICC for all measurements ranged from 0.66 to 0.97, and indicated moderate to good reliability and reproducibility of all soft tissue landmarks.

Statistical analysis

The proportion index values were calculated by comparing the UCLP subjects with the control subjects. The proportion indices were converted to z scores by using the following formula: \( z = (x - \mu) / \sigma \); where \( z \) is z scores; \( x \) is UCLP patient’s facial index; \( \mu \) = mean of the control subjects; and \( \sigma \) = SD of the control subjects.

Z scores are a statistical measurement to see how far the evaluated data deviate from the mean of the normal measurements. The anthropometric categories for z scores were classified based on the normal values: harmony (−1.0 to +1.0) and disharmony (−2.0 to −2.0 or +1.0 to +2.0). Disproportions were: mildly subnormal (−2.0 to −2.50), moderately subnormal (−2.51 to −3.00), severely subnormal (−3.01
to $-9.99$), mildly supernormal ($+2.01$ to $+2.50$), moderately supernormal ($+2.51$ to $+3.00$), and severely supernormal ($+3.01$ to $+9.99$) (Fig. 2).\textsuperscript{1,12,16,17}

We used the independent sample $t$ test to assess the significance of differences between the indices in control patients and those with UCLP. Probabilities of less than 0.05 were accepted as significant, and analysis was aided by SPSS (version 17.0, SPSS Inc., Chicago, IL, USA).

### Results

Table 2 shows differences in the proportion indices with mean differences, probabilities, $z$ scores, and the categories for $z$ scores between control patients and those with CLP. From the 11 calculated indices, 4 showed normal harmony and 5 normal disharmony. The normal harmony was shown in the upper face, fissures of the right and left eyes, and the skin portions of the upper lip, and normal disharmony was shown in the morphological facial area, lower face, intercanthal distance, protrusion of the nasal tip, and height of the upper vermilion. Two indices were disproportionate: nasal index (which was severely supernormal) and upper lip index (which was moderately supernormal). The probabilities also indicated that there were significant differences between normal faces and those with UCLP in 7 indices: morphological facial area, lower face, intercanthal distance, nasal, protrusion of the nasal tip, upper lip, and height of the upper vermilion indices ($p = 0.001, 0.001, 0.001, 0.001, 0.007, 0.001, 0.044$).

### Discussion

We have examined the quantitative values of measurements that use proportion indices in the craniofacial regions in patients with repaired, non-syndromic complete UCLP using...
non-invasive systems of the 3-dimensional stereophotogrammetry technology, the VECTRA-3D dual module system for full-face imaging (Canfield Scientific Inc., Fairfield, NJ, USA). Data were compared with those collected from patients without clefts of the same age and ethnic group by using probabilities and z scores. The VECTRA-3D facial scanner has proved to be a reliable, accurate, and repeatable system for recording and measuring facial soft tissue morphology (Asi SM et al. Validity and reliability of data acquisition using Vectra 3D compared to direct method. Paper presented at biomedical engineering and sciences (IECBS) conference, 2012 December). The sample age group, 18–25 years old, was selected to assess the effects of primary and palatal repair after major growth had ceased.

We found significant differences between the 2 groups of patients in 7 out of 11 craniofacial proportion indices that were assessed (p = 0.001–0.044). In clinical practice, differences are recorded in mm. To our knowledge none of the previous studies has suggested thresholds for clinically relevant differences expressed in mm. We therefore propose that 5 mm is an appropriate threshold to indicate clinical relevance. This means that a clinician can detect any abnormalities on the face of a patient with UCLP only if the difference is 5 mm or more than that of normal patients. We have further shown that of the 7 craniofacial proportion indices that were significantly different, only 5 were clinically relevant (mean difference 5 mm or more). As expected, the most affected craniofacial regions were the nose (nasal and nasal tip) and orolabial (upper lip and height of upper vermillion). “Morphological face” is the

Table 1
Linear measurements and proportional indices derived from anthropometric soft tissue landmarks.

<table>
<thead>
<tr>
<th>Regions</th>
<th>Linear measurements</th>
<th>Proportional indices</th>
</tr>
</thead>
<tbody>
<tr>
<td>Face</td>
<td>Width of the face (zy-zy)</td>
<td>Morphological facial index = (Width of the face (zy-zy) / Height of the upper face (n-gn)) × 100</td>
</tr>
<tr>
<td></td>
<td>Morphological facial height (n-gn)</td>
<td>Upper face index = (Width of the face (zy-zy) / Morphological facial height (n-gn)) × 100</td>
</tr>
<tr>
<td></td>
<td>Height of the upper face (n-sto)</td>
<td>Lower face index = (Width of the face (zy-zy) / Morphological facial height (n-gn)) × 100</td>
</tr>
<tr>
<td>Orbits</td>
<td>Intercanthal width (enR-enL)</td>
<td>Intercanthal index = (Intercanthal width (enR-enL) / Intercanthal width (exR-exL)) × 100</td>
</tr>
<tr>
<td></td>
<td>Biocorlar width (exR-exL)</td>
<td>Right eye fissure index = (Right eye fissure length (psR-piR) / Left eye fissure length (psL-piL)) × 100</td>
</tr>
<tr>
<td></td>
<td>Right eye fissure length (exR-exL)</td>
<td>Left eye fissure index = (Left eye fissure length (psL-piL) / Left eye fissure length (exL-enL)) × 100</td>
</tr>
<tr>
<td>Nose</td>
<td>Height of the nose (n-sn)</td>
<td>Nasal index = (Width of the nose (al-al) / Height of the nose (n-sn)) × 100</td>
</tr>
<tr>
<td></td>
<td>Width of the nose (al-al)</td>
<td>Nasal tip protrusion width index = (Nasal tip protrusion (sn-prn) / Width of the nose (al-al)) × 100</td>
</tr>
<tr>
<td></td>
<td>Nasal tip protrusion (sn-prn)</td>
<td>Intercanthal index = (Intercanthal width (exR-exL) / Biocorlar width (exL-enL)) × 100</td>
</tr>
<tr>
<td>Orolabial</td>
<td>Width of the mouth (ch-ch)</td>
<td>Upper lip width index = (Height of the upper lip (un-sto) / Height of the mouth (ch-ch)) × 100</td>
</tr>
<tr>
<td></td>
<td>Height of the upper lip (sn-sto)</td>
<td>Skin portion upper lip index = (Height of the upper lip (sn-sto) / Height of the upper lip (al-al)) × 100</td>
</tr>
<tr>
<td></td>
<td>Vermilion height of the upper lip (ls-sto)</td>
<td>Upper vermilion height index = (Vermilion height of the upper lip (ls-sto) / Height of the upper lip (al-al)) × 100</td>
</tr>
</tbody>
</table>

Table 2
Differences in proportion indices between normal patients and those with cleft lip and palate. Data are mean (SD) (mm) except where otherwise stated.

<table>
<thead>
<tr>
<th>Region and proportion index</th>
<th>Normal (n = 100)</th>
<th>Cleft lip and palate (n = 15)</th>
<th>Mean difference (mm)</th>
<th>p Value</th>
<th>Z score</th>
<th>Categories for z scores</th>
</tr>
</thead>
<tbody>
<tr>
<td>Face</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Morphological face</td>
<td>92.17 (9.37)</td>
<td>105.45 (10.90)</td>
<td>13.28</td>
<td>0.001</td>
<td>+1.42</td>
<td>Disharmony</td>
</tr>
<tr>
<td>Upper face</td>
<td>61.3 (6.94)</td>
<td>63.46 (4.29)</td>
<td>2.16</td>
<td>0.29</td>
<td>+0.31</td>
<td>Harmony</td>
</tr>
<tr>
<td>Lower face</td>
<td>53.55 (2.68)</td>
<td>58.21 (3.03)</td>
<td>4.67</td>
<td>0.001</td>
<td>+1.74</td>
<td>Disharmony</td>
</tr>
<tr>
<td>Orbit</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Intercanthal distance</td>
<td>32.76 (2.5)</td>
<td>36.63 (2.8)</td>
<td>3.88</td>
<td>0.001</td>
<td>+1.55</td>
<td>Disharmony</td>
</tr>
<tr>
<td>Right eye fissure</td>
<td>36.63 (5.14)</td>
<td>37.61 (5.23)</td>
<td>0.68</td>
<td>0.69</td>
<td>+0.19</td>
<td>Harmony</td>
</tr>
<tr>
<td>Left eye fissure</td>
<td>36.93 (5.15)</td>
<td>38.14 (4.66)</td>
<td>1.22</td>
<td>0.46</td>
<td>+0.23</td>
<td>Harmony</td>
</tr>
<tr>
<td>Nose</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nasal</td>
<td>67.67 (7.2)</td>
<td>90.59 (14.8)</td>
<td>22.92</td>
<td>0.001</td>
<td>+3.18</td>
<td>Severely supernormal</td>
</tr>
<tr>
<td>Width of protrusion of nasal tip</td>
<td>43.4 (4.59)</td>
<td>38.4 (6.2)</td>
<td>5.0</td>
<td>0.007</td>
<td>-1.09</td>
<td>Disharmony</td>
</tr>
<tr>
<td>Orolabial</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Upper lip</td>
<td>43.6 (3.97)</td>
<td>54.56 (6.67)</td>
<td>10.95</td>
<td>0.001</td>
<td>+2.76</td>
<td>Moderately supernormal</td>
</tr>
<tr>
<td>Skin portion of upper lip</td>
<td>63.49 (7.13)</td>
<td>60.53 (15.21)</td>
<td>2.95</td>
<td>0.42</td>
<td>-0.42</td>
<td>Harmony</td>
</tr>
<tr>
<td>Height of upper vermilion</td>
<td>85.97 (11.7)</td>
<td>74 (24.1)</td>
<td>11.96</td>
<td>0.04</td>
<td>-1.02</td>
<td>Disharmony</td>
</tr>
</tbody>
</table>
only index in the facial region that was found to be clinically relevant, and there was none from the orbital region.

Based on the results of analysis of $z$ scores, normal values were present in the proportion index of morphological face, upper and lower face, intercanthal distance, fissure of the right and left eye, protrusion width of the nasal tip, skin portion of the upper lip and height of the upper vermilion (Table 2). We found harmony in the upper face, fissures of the right and left eye, and skin portion upper lip indices. This was in all regions except the nose. Disproportions were present in the nose region where “nasal” was severely supernormal and the upper lip was moderately supernormal. Most of the patients with UCLP had higher mean $z$ scores, which meant that generally patients with UCLP had larger faces than normal control subjects. The higher mean $z$ scores for both nasal and upper lip indices showed that the nose and upper lip were wider, larger, or flatter in patients with UCLP than in control patients.

The results of the present investigation generally support the findings of earlier studies in which there were significant differences in the nasal and orolabial regions of patients with UCLP compared with those of subjects without clefts. 1,4,6,7,12,19-23 Zreaqat et al. 19 published a comparative cross-sectional study in which they assessed the facial surface dimensions of a group of Malay children with UCLP using 3-dimensional analysis analysis. Their results showed significant dimensional differences mainly in the nasolabial region between the Malay children with UCLP and the control group. The width of the alar base and alar base root were increased, as was the width of nose base:mouth ratio in the group with UCLP. Although our sample’s ethnicity was similar to that in their study, they used mainly linear measurements and the sample age was 8–10 years old. They explained that their finding might have resulted from failure to restore the loss of the medial insertion points of the facial muscle during primary operations, which flattened and elongated the ala and resulted in an increase in the horizontal nasal dimension.

Farkas et al. 12 were among the first authors who compared the morphology of the patients with CLP with that of normal controls using proportion indices, and reported disproportions in the soft tissue that included a wide nose, a small protrusion of the nasal tip, and a short or long columella. Here the mild, moderate, and severe scales were introduced to grade the disproportions and measurements that contributing to them. In the present study, patients with UCLP inhibited supernormal measurements, which confirms the results of Farkas et al. 12 who found, in their group with unilateral clefts, that supernormal measurements were much more common than subnormal ones and that the nasal index was in the severely supernormal category. In this particular case they had used the traditional technique of direct measurement, whereas we used the non-invasive systems of the 3-dimensional stereophotogrammetry technology. Nevertheless, both studies found that patients with UCLP tended to have more problems in the lower part of the face (nasal and orolabial).

A more recent study used 3-dimensional technology, and measurements were made by dense correspondence surface techniques. In computer vision and graphics, establishment of the correspondence of 3-dimensional points between surfaces (such as on the human face) has many important applications that involve 3-dimensional surface reconstruction from images, and comparisons that rely on the representation of shapes. The accuracy of the correlation is contingent on the recording process, and many different approaches (such as genetic algorithms) have been proposed to improve it. It has been agreed that it is a good way to solve complex optimisation problems of deformable surface meshes (Zhu L, Lee WS. Facial expression via genetic algorithms. Paper presented at 19th annual conference on computer animation and social agents (CASA), Geneva, 2006 July). Bugaighis et al. 22 used it when they superimposed each of the “average” faces of the group with clefts on the control “average” face using surface-based recording with 3dMD software (3dMD Technologies, Guildford, UK), where a colour-coded image displays the subjective differences between the 2 surfaces. 22 It was a prospective cross-sectional, case-controlled, morphometric study that involved 40 patients with UCLP and 80 white controls matched for age and sex. Although the study used a more sophisticated measuring method (statistical shape analysis), they also found that the differences were mainly in the nasolabial region. The groups with UCLP had wider, more asymmetrical noses, with flattening at the dome of the nasal crura and appreciable flattening of the furrow of the philtrum.
It has been postulated that the growth-retardant effect of operation for CLP can be seen transversely as high as the orbital level. We found that there was a significant difference (p = 0.001), although it was not clinically relevant (mean difference 3.88 mm) in the intercanthal proportion index between the groups. Z scores indicated definite disharmony, which meant that in the group with UCLP the intercanthal dimension was larger than in the normal group. This contradicts previous reports that showed no significant difference at the level of the eye in ethnic Malays, and significantly narrower intercanthal width in white patients with UCLP compared with controls. The differences might be because the patients with UCLP were younger in both studies, and of different ethnicity in one of them.

Within limitations, and based on the results of the present study, there were significant and clinically relevant differences mainly in the nasolabial area between the 2 groups of subjects. Certain surgical techniques used by many surgeons from different specialties during the primary or even secondary operations might have contributed to these findings. It is crucial to classify the level of these disproportions so that we can identify the defective elements to be repaired. This is also important in assessment of the degree of success of the corrective surgery or in making decisions about further planning. The use of the anthropometric non-invasive systems of the 3-dimensional stereophotogrammetry technology has potential in the evaluation of the outcome of the operation and planning of reconstruction if needed.

Conflict of interests

We have no conflict of interest that we should disclose.

Acknowledgments

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