Facial shape and asymmetry in 5-year-old children with repaired unilateral cleft lip and/or palate: an exploratory study using laser scanning


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SUMMARY To investigate the feasibility of facial laser scanning in pre-school children and to demonstrate landmark-independent three-dimensional (3D) analyses for assessment of facial deformity in 5-year-old children with repaired non-syndromic unilateral cleft lip and/or cleft palate (UCL/P).

Faces of twelve 5-year-old children with UCL/P (recruited from university hospitals in Cardiff and Swansea, UK) and 35 age-matched healthy children (recruited from a primary school in Cardiff) were laser scanned. Cleft deformity was assessed by comparing individual faces against the age and gender-matched average face of healthy children. Facial asymmetry was quantified by comparing original faces with their mirror images.

All facial scans had good quality. In a group of six children with isolated cleft palate coincidence with the average norm ranged from 18.8 to 26.4 per cent. There was no statistically significant difference in facial asymmetry when compared with healthy children ($P > 0.05$). In a group of six children with UCL with or without cleft palate coincidence with the average norm ranged from 14.8 to 29.8 per cent. Forehead, midface and mandibular deficiencies were a consistent finding, ranging from 4 to 10 mm. The amount of 3D facial asymmetry was higher in this group ($P < 0.05$).

Facial laser scanning can be a suitable method for 3D assessment of facial morphology in pre-school children, provided children are well prepared. Landmark-independent methods of 3D analyses can contribute to understanding and quantification of facial soft tissue cleft deformity and be useful in clinical practice.

Introduction

Recent innovations in non-invasive three-dimensional (3D) imaging technology provide tools for comprehensive assessment of facial surface anatomy, which is of particular importance in patients with dentofacial deformity. In the majority of studies performed on patients with cleft lip and/or cleft palate (CL/P) stereophotogrammetry has been the method of choice for capturing 3D facial data (Bilwatsch et al., 2006; Devlin et al., 2007; Bugsaghis et al., 2010; van Loon et al., 2010, 2011). This is mainly due to fast capture time, simplicity of application, reliable identification of landmarks on photorealistic life-like models, and reliable storing and archiving of data (Ayoub et al., 2003, Devlin et al., 2007, Heike et al., 2010).

Another non-invasive technology, which has been accepted by orthodontists, maxillofacial and plastic surgeons, is laser surface scanning (Duffy et al., 2000; Da Silveira et al., 2003; Moss, 2006; Kau et al., 2007; Schwener-Zimmerer et al., 2008; Kau and Richmond, 2010; Kau et al., 2010; Toma et al., 2008, 2009, 2011).

It has been proved that this method is valid and reliable (Kau et al., 2004, 2005, 2006a; Kovacs et al., 2006). However, some authors have suggested that it might not be suitable for use in the paediatric population due to the relatively long capture time (Al-Omari et al., 2005; Devlin et al., 2007). Till date, no formal reliability study has been carried out in young children, although laser scanning has been applied in 5–6 year olds to evaluate the results of an early crossbite correction on the face (Primožič et al., 2005, 2009, 2012) and to analyse facial characteristics of children with class III malocclusion (Kmetka et al., 2012). Interest in the application of this technique lies in the fact that laser scanning devices are less expensive, portable, and produce very accurate 3D facial models (Kau and Richmond, 2010a). It is crucial to obtain 3D normative data for patients with cleft lip and palate using a non-invasive technique, which is cost-effective, accurate, reliable, and easy to use. The importance of an early assessment of patient’s face lies in the determination of the residual treatment need and planning.
of future health care measures. Therefore, the aims of this exploratory study were: 1) to investigate the feasibility of facial laser scanning in pre-school children and 2) to demonstrate landmark-independent 3D analyses of facial shape and asymmetry in 5-year-old children with repaired non-syndromic unilateral cleft lip and/or cleft palate (UCL/P).

Materials and methods

Sample
A total of 12 children [eight males and four females, average age 5.3 (standard deviation [SD] 0.5) years] with repaired non-syndromic UCL/P were voluntarily recruited from two university hospitals in Cardiff and Swansea, UK. Three children had complete UCL (two males and one female), six children had isolated cleft palate (three males and three females), and three males had cleft lip and palate. All cleft lips were located on the left side. Primary surgical closure of the lip was performed at approximately 3 months of age using Millard technique with vomer flap. Palatal surgery was performed between 9 and 12 months of age using von Langenbeck technique with a limited muscle dissection. None of the patients had any secondary surgery and all were white British. A total of 35 healthy children [17 males, 18 females, average age 5.5 (SD 0.5) years] were voluntarily recruited from a reception year at a primary school in Cardiff. These children met the following criteria: white British, absence of facial trauma, surgical intervention, dysmorphology, and marked asymmetry (as assessed by two examiners) and consent signed by the parent or legal guardian. Ethical approval was obtained from the relevant ethics committee, the dental school committee and the head teacher.

Image capture
Two Vivid 900 digitizers (Konica Minolta Sensing Europe, Milton Keynes, UK) were used to scan faces. These devices emit an eye-safe Class 1 laser according to US Food and Drug Administration and Class 2 laser according to the International Electrotechnical Commission (Konica Minolta Sensing Inc., Konica Minolta Instruction Manual, 2001–03). Medium range lenses with a focal length of 14.5 mm were used (Kau et al., 2004, 2005). The children were invited in groups of four and given an explanatory talk and a demonstration prior to scanning. Each child sat on a self-adjustable stool and was asked to look directly at the investigator seat ed 135 cm in front of them, between the scanning devices. Neutral facial expression and a natural head position were adopted (Kau and Richmond, 2008). The total scan time was approximately 8 seconds. If it was perceived that the child smiled, changed facial expression, or moved between the scans, the procedure was repeated. One raw data set, consisting of one left and one right laser scan, was captured. The child was assisted in remaining motionless by stimulation of the senses of sight and hearing. The investigator seated directly in front of the child and counted aloud from 1 to 8 and held the corresponding number of fingers aloft as a visual aid. This encouraged the child as they were made aware of the amount of time they were required to keep still.

Image processing
Processing of facial scans was performed in Rapidform 2006 (INUS Technology Inc., Seoul, South Korea). Using an in-house developed subroutine, unwanted extraneous data (neck, ears, hair, and clothing) was removed and images were smoothed taking care of their shape and volume (Zhurov et al., 2005). Prior to merging facial halves, the registration result was checked to verify the scanning accuracy (Figure 1). A previous study on reliability of laser scanning in a group of 11 year olds showed that at least 90 per cent of match in the overlap area of facial halves, with an error up to 0.75 mm, is clinically acceptable (Kau et al., 2004). The same principle was accepted in this study. After merging, three landmarks were manually identified on the faces: endocanthion left and right (inner corners of the eyes) and pogonion (the most prominent point on the chin) to assist standardisation (Kau and Richmond, 2010). Mirror face was automatically created, and the structure consisting of original and mirror faces (best-fit registered) served as a basis for defining coronal and sagittal planes. Coronal plane (xy) was determined by the cylinder, which fitted all data points of the original-mirror face structure. Sagittal plane (yz) was determined as the symmetry plane of the original-mirror face structure. Transverse plane (xz) was perpendicular to previous two planes (Kau and Richmond, 2010). The origin of this co-ordinate system was the mid-endocanthion point (a point halfway between inner corners of the eyes), as previous research has shown that its position changes least with time (Farkas, 1994) and across a sample of 350 adolescent faces (Toma et al., 2008, 2009; Kau and Richmond, 2010).

Facial shape assessment
Surface-based average faces were constructed separately for males \(n = 17\) and females \(n = 18\) in the group of healthy children (Figure 2). Prior to averaging, faces were fitted into the same reference frame, as explained above. Different averaging methods have been described in the literature: straightforward pointwise averaging in the z-direction; averaging in the radial direction of the average facial cylinder; averaging in the radial direction of the average facial sphere; and averaging in the locally normal direction to a template face (Kau and Richmond, 2010). In this study, male and female facial averages were obtained by calculating pointwise mean co-ordinates in the direction nearly perpendicular to all faces. This approach was implemented using a template face (randomly chosen from the sample) and calculating means in the locally normal direction to that template. The resulting average face was further used as a new template and the averaging was repeated. Three
iterations were performed in order to get average faces with good accuracy (Kau and Richmond, 2010).

Individual faces of children with clefts were compared with the average male and female faces of healthy children, by superimposing them on the mid-endocanthion point. Comparisons were performed separately in two groups: group 1 consisting of six children (three males and three females) with repaired isolated cleft palate (Figure 3) and group 2 consisting of six children (five males and one female) with repaired UCL with or without cleft palate (Figure 4). Signed colour maps were analysed by direct observations and using data provided by accompanying histograms (percentage of coincidence between the faces and difference in millimetres for any given facial area). Shades of blue represented deficient areas (negative difference) on the cleft faces when they were compared with the norm (age and gender-matched average face of healthy children). Shades of red represented areas of the face where soft tissue of the cleft face was more prominent than the norm (positive difference).

**Facial asymmetry assessment**

The original and the mirror faces were superimposed (best-fit registered) in order to quantify the amount of 3D facial...
asymmetry (Figure 5). This method has been previously applied in adolescents (Djordjevic et al., 2011a,b). The average distance between the two facial surfaces was calculated, as well as the percentage of coincidence within 0.5 mm of tolerance (arbitrarily chosen for asymmetry assessment). The amount of facial asymmetry increases with greater average distance between the two faces (original and mirror one) and lower percentage of coincidence. The face was divided into four regions by three horizontal planes: the first plane connecting inner corners of the eyes, the second plane passing through the point subnasale (at the base of the nose) and normal to mid-sagittal plane, and the third plane connecting the corners of the lips. This was done in order to assess asymmetry of the nose and upper lip with surrounding tissue separately, as previously suggested in the literature (Hood et al., 2003).

Statistical analysis
The quality of the scans was analysed using descriptive statistics. Histograms, Q–Q plots, and Shapiro–Wilk test were used to check the distribution of facial asymmetry parameters. Because the distribution was not normal and the sample was small, transformation of the data was not attempted. The data are presented as median and interquartile range (25th percentile, 75th percentile). The non-parametric Mann–Whitney U-test was used for comparisons between the two cleft groups and healthy children.
Facial asymmetry was not analysed separately for males and females due to the small sample size. Due to the exploratory nature of the study, Bonferroni correction for multiple testing was not considered, and a $P$ value less than 0.05 was deemed statistically significant. Statistical analyses were performed in SPSS version 17.0 (SPSS Inc., Chicago, Illinois, USA).

**Results**

The average distance between the right and left facial scans in the overlap area was 0.25 (SD 0.06) mm, and the coincidence was 95.1 (SD 2.9) per cent, within a tolerance level of 0.75 mm. As quality criteria were satisfied, all scans could be processed, merged, and analysed.

Surface-based average faces of males and females from the control group were superimposed using a best-fit registration. Gender differences in facial shape are graphically presented on a colour map (Figure 2). Females tend to have more prominent eyes and cheeks, with a maximum difference of 2.5 mm. Males tend to have more prominent upper part of the forehead, bridge of the nose, and the lips, with a maximum difference of 2.0 mm. Coincidence of the two average faces was 43.7 per cent, within 0.5 mm of tolerance.

The superimpositions highlight the differences between facial soft tissues of children with repaired isolated cleft

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**Figure 4** Faces of six children (five males and one female) with repaired unilateral cleft lip with or without cleft palate were compared with gender and age-matched average faces (presented in Figure 2). Transparent profile views and signed colour maps with histograms are shown. Individual faces are coloured in grey, average male face in blue, and average female face in violet. Shades of red and blue on the colour maps represent parts of individual face, which are more prominent (positive difference) or less prominent (negative difference), respectively, when compared with the norm. Grey areas indicate the coincidence of the two faces within 0.5 mm tolerance. The scale is standardised, with a range from −10 to 10 mm and divisions of 2 mm.
Figure 5  The method for three-dimensional assessment of facial asymmetry. The original and the mirror faces (upper left) were superimposed (best-fit registered) in Rapidform 2006 (Inus Technology Inc., Seoul, South Korea). The signed colour map (upper middle) represents deviations between two facial surfaces for the whole face. The scale is standardised (−4.5 to 4.5 mm), and the shades of blue and red represent asymmetric areas. Further analysis of asymmetry included four different regions, determined by three planes. The first region (R1 in Table 1) represents the area above the line which connects the inner corners of the eyes (upper right); the second region (R2) occupies the area between the line which connects the inner corners of the eyes and the line which passes through the point subnasale (at the base of the nose) and is normal to mid-sagittal plane (lower left); the third region (R3) is determined by the line passing through the point subnasale and the line which connects corners of the mouth (lower middle); and the fourth region (R4) is located below the line which connects corners of the mouth (lower right). The average distance between the two surfaces and the percentage of their coincidence (within 0.5 mm) were measured using in-house developed subroutine for Rapidform 2006.

Table 1  Three-dimensional facial asymmetry of children with repaired isolated cleft palate, unilateral cleft lip with or without cleft palate, and healthy control group.

<table>
<thead>
<tr>
<th>Parameters</th>
<th>ICP group (n = 6)</th>
<th>UCL ± P group (n = 6)</th>
<th>Control group (n = 35)</th>
<th>ICP versus control</th>
<th>UCL ± P versus control</th>
</tr>
</thead>
<tbody>
<tr>
<td>Whole face</td>
<td></td>
<td></td>
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<td></td>
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<tr>
<td>Average distance (mm)</td>
<td>0.52 (0.49, 0.66)</td>
<td>0.88 (0.64, 0.95)</td>
<td>0.49 (0.40, 0.59)</td>
<td>0.356 (NS)</td>
<td>0.002*</td>
</tr>
<tr>
<td>Coincidence (%)</td>
<td>63.3 (45.4, 69.2)</td>
<td>48.4 (45.8, 52.8)</td>
<td>65.5 (53.6, 70.0)</td>
<td>0.733 (NS)</td>
<td>0.006*</td>
</tr>
<tr>
<td>R1</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Average distance (mm)</td>
<td>0.61 (0.36, 0.74)</td>
<td>0.44 (0.42, 0.50)</td>
<td>0.44 (0.36, 0.56)</td>
<td>0.482 (NS)</td>
<td>0.679 (NS)</td>
</tr>
<tr>
<td>Coincidence (%)</td>
<td>57.7 (48.4, 74.6)</td>
<td>65.2 (63.3, 70.4)</td>
<td>68.7 (51.9, 75.6)</td>
<td>0.760 (NS)</td>
<td>0.986 (NS)</td>
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<tr>
<td>R2</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Average distance (mm)</td>
<td>0.42 (0.34, 0.62)</td>
<td>0.76 (0.56, 0.89)</td>
<td>0.39 (0.31, 0.50)</td>
<td>0.528 (NS)</td>
<td>0.001*</td>
</tr>
<tr>
<td>Coincidence (%)</td>
<td>73.9 (49.7, 80.6)</td>
<td>47.9 (44.0, 53.0)</td>
<td>73.7 (60.8, 82.7)</td>
<td>0.733 (NS)</td>
<td>0.001*</td>
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<tr>
<td>R3</td>
<td></td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Average distance (mm)</td>
<td>0.53 (0.46, 0.62)</td>
<td>0.89 (0.72, 1.43)</td>
<td>0.46 (0.34, 0.62)</td>
<td>0.552 (NS)</td>
<td>0.002*</td>
</tr>
<tr>
<td>Coincidence (%)</td>
<td>53.4 (50.8, 59.7)</td>
<td>36.3 (27.3, 40.5)</td>
<td>62.8 (49.7, 77.7)</td>
<td>0.679 (NS)</td>
<td>0.007*</td>
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<tr>
<td>R4</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Average distance (mm)</td>
<td>0.72 (0.51, 1.00)</td>
<td>1.45 (0.90, 2.15)</td>
<td>0.68 (0.45, 0.90)</td>
<td>0.679 (NS)</td>
<td>0.023*</td>
</tr>
<tr>
<td>Coincidence (%)</td>
<td>36.3 (22.9, 50.8)</td>
<td>14.1 (9.6, 26.4)</td>
<td>41.3 (29.2, 60.8)</td>
<td>0.505 (NS)</td>
<td>0.011*</td>
</tr>
</tbody>
</table>

ICP, isolated cleft palate; NS, not statistically significant; UCL ± P, unilateral cleft lip with or without cleft palate.
Medians and interquartile range (25th and 75th percentile) are presented.
Mann–Whitney U-test was performed.
For an explanation of regions R1, 2, 3, and 4, refer to the text and Figure 5.
*P < 0.05.
palate and their healthy peers (Figure 3). The percentage of coincidence between individual faces and age and gender-matched average faces (norms) ranged from 18.8 to 26.4 per cent, for the tolerance level 0.5 mm. Both positive and negative differences were noted in this cleft group, ranging from 2 to 7 mm. Less protrusive upper and lower lips and philtrum were noted in three children. Half of the group had positive difference in the forehead region and the other half had a negative difference. One child had more protrusive facial soft tissue overall. Varying degree of soft tissue deficiency at the tip and/or bridge of the nose could also be demonstrated.

The superimpositions for the second group of six children with repaired UCL with or without cleft palate are presented in Figure 4. The percentage of coincidence between individual faces and age and gender-matched norms ranged from 14.8 to 29.8 per cent, for the tolerance level 0.5 mm. Similar to previous group, both positive and negative differences were noted, ranging from 4 to 10 mm. Retrusion of the forehead, midface, and mandible were consistent findings in this group. Two children had incompetent lips, which affected the amount and direction of difference on the colour maps.

Preliminary findings on facial asymmetry (Table 1) did not reveal any statistically significant difference between children with repaired isolated cleft palate and healthy children ($P > 0.05$). Statistically significant difference was noted in the group of children with repaired UCL with or without cleft palate ($P > 0.05$). In these children, the amount of facial asymmetry was higher in all measured regions, except the forehead.

**Discussion**

Various 2D and 3D methods have been suggested for analysis of cleft faces and there is still a need for an internationally agreed objective method of assessment (Al-Omari et al., 2005). The first aim of the study was to explore the possibility of using laser scanning as a non-invasive 3D method for capturing facial soft tissue morphology in pre-school children. It has been argued that its main disadvantage is a long capture time (Al-Omari et al., 2005; Devlin et al., 2007). Although good cooperation with younger children might be quite challenging, in the present study, it was accomplished mainly due to an explanatory talk prior to the procedure and the use of visual and audio signals during it.

The results indicate adequate scanning accuracy, comparable to previous studies performed on adolescents and adults (Kau et al., 2004, 2005; Toma et al., 2008, 2009). If operators cannot achieve good cooperation with individuals and scanning quality is not adequate using two devices, it is suggested one device be used, positioned directly in front of an individual’s face. In this way, capturing time can be effectively reduced to approximately 3 seconds. To some extent, it would decrease lateral aspects of the face presented on a 3D model, but it would still be sufficient for the analysis of cleft deformity.

Another aim of this study was to demonstrate landmark-independent 3D analyses for assessment of facial deformity in children with UCL/P. Instead of relying on relatively small number of landmarks and their respective 3D co-ordinates, a landmark-independent, surface-based method is presented. The analyses were entirely based on comparisons of facial surfaces, making the maximum use of tens of thousands of data points, which were captured by the laser scanning device. Surface-based average faces have been used to investigate facial growth, to assess orthodontic and orthognathic treatment outcomes, to determine gender and ethnic differences among populations, and to delineate between some craniofacial syndromes (Kau et al., 2006b, 2010a,b; Hammond, 2007; Kau and Richmond, 2008; Toma et al., 2008). However, they have been rarely applied in the context of facial cleft analysis (Duffy et al., 2000).

In this study, the average faces were constructed for males and females separately, as superimpositions indicated that there was a gender difference in facial shape in the group of healthy children. This is in agreement with the study performed by Moss (2006). Individual faces of six children with repaired isolated cleft palate and six children with repaired UCL with or without palate were compared with these average faces, which served as the age and gender-specific norms. Midface deficiency was more consistent finding in the latter group. The following explanations for this deformity have been suggested: the restriction of maxillary growth due to primary surgery, intrinsic growth impairment, or a combination of these two factors (Mars, 2004; Devlin et al., 2007). In addition to differences in the cleft region, an interesting finding worth further exploration is the less prominent forehead in some children with clefts. This coincides with the findings of Duffy et al. (2000) in 8–11 year olds with repaired UCLP.

This was an exploratory study performed on a small sample of 5 year olds with UCL/P, who were recruited from two different centres. The main idea was to demonstrate the possibilities of 3D quantification of soft tissue deformity using landmark-independent approach. Hence, more studies with larger samples and good control of possible confounding factors (such as general health status, body mass index, timing and type of primary surgery) will be needed to fully characterise facial deformity in this young group of patients.

Visual representation of shape differences by a colour map and its quantification on an accompanying histogram might form a basis for an assessment of residual treatment need and adequate treatment planning. If the surgical goal is to achieve a facial shape close to that of normal children, then normal range of facial shape should be used. In this study, no attempt was made to select the control group of healthy children on the basis of their facial skeletal
parameters or growth pattern. There were two reasons behind this decision: first, healthy children were randomly selected from a local school and therefore no exposure to radiation would be ethically justified; second, it has been shown on a sample of 4747 adolescents that facial shape of normal individuals can be considered a multidimensional statistical continuum and that average faces can be constructed to reflect the differences in principal components of facial shape, such as height, width, and nose prominence (Toma et al., 2011). Therefore, sample of healthy children in this study is a realistic representation of range of facial morphology in this age group. Previous studies showed that there might be a considerable overlap in facial morphology of some cleft deformities and normal individuals (Yamada et al., 2002; Bugaighis et al., 2010) and that should also be investigated further using suggested methodology.

Residual nasolabial asymmetry after primary reconstructive surgery in children with UCL/P has been confirmed in previous studies (Ras et al., 1994; Bilwatsch et al., 2006; Devlin et al., 2007; Schwenzer-Zimmerer et al., 2008; Stauber et al., 2008; Bugaighis et al., 2010). Because it can cause serious aesthetic concerns to patients and clinicians (Bugaighis et al., 2010; Fudalej et al., 2012), it demands an objective 3D assessment. Even aesthetically pleasing faces exhibit facial asymmetry to some extent. Therefore, a range of normal asymmetry scores have to be determined in a reference population before a direct comparison with cleft deformity can be made. As in facial shape assessment, most studies rely on landmark-dependent methods, measurements of linear distances, angles, or geometric morphometrics to perform Procrustes registration of landmark configurations (original and reflected ones) (Hood et al., 2003; Hajeer et al., 2004; Devlin et al., 2007; Stauber et al., 2008; van Loon et al., 2011).

In this study, overall facial asymmetry was assessed, as well as the contribution of four different facial regions. The method was based on the comparison of the original and mirror images. Colour maps provided both qualitative and quantitative data on facial asymmetry. Preliminary findings did not show any statistically significant difference in the amount of facial asymmetry between children with repaired isolated cleft palate and their healthy peers. However, in the second group of children with UCL with or without cleft palate, significant differences were found. These children had higher amount of asymmetry in all facial regions measured, except the upper part (forehead). No definite conclusions can be drawn at this point due to small sample sizes. However, the suggested method can be used in everyday practice, as it can be easily understood and interpreted (both by clinicians and patients). In addition, this approach would allow us to compare different groups of cleft patients (in the context of timing and type of surgery) or the same group of cleft patients before and after treatment.

A recent study found that the standard of care for children with cleft lip and palate in the United Kingdom has improved in the last 15 years, with reduction in the number of centres operating on these children from 57 in 1998 to 11 in 2011 (Sandy et al., 2012). With fewer centres and a culture of regularly inviting patients and families to review clinics at age 5 years, early assessment of facial deformity after primary surgery will be easier to perform on a much larger sample. Therefore, a more comprehensive and consistent analysis of treatment outcome in general, including 3D facial analysis, could be performed. It is hoped that some preliminary observations made in this study can contribute to further trials.

Conclusion

The following conclusions can be drawn from this exploratory study: 1) Laser scanning can be a suitable method for 3D assessment of facial morphology in pre-school children, provided children are well prepared. 2) Landmark-independent 3D analyses can contribute to understanding and quantification of facial soft tissue cleft deformity and be useful in clinical practice.

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