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Three-dimensional facial morphology following surgical repair of unilateral cleft lip and palate in patients after nasoalveolar molding

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Structured Abstract
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Objective – To evaluate three-dimensional (3D) facial morphology in patients surgically corrected for unilateral cleft lip and palate (UCLP) following pre-surgical nasoalveolar molding (NAM).
Design – Prospective, longitudinal study. Digital stereophotogrammetry was used to capture 3D facial images, and *x*, *y*, and *z* coordinates of five landmarks were digitized to compute mean morphologies. The sample comprised 15 patients with left UCLP and 10 matched control subjects. Facial form differences at age 37 weeks, using principal components analysis and finite-element scaling analysis (FESA) were assessed.
Results – Using the first two principal components, which accounted for 63% of the total shape-change, UCLP and control groups showed similar distributions in the modal space ($p > 0.05$). For the UCLP group, the mean 3D facial form was smaller and less protrusive when superimposed on the non-cleft mean. Using FESA, reductions in facial volume were found in the UCLP group, involving the columella (29%), labial tubercle (51%), lower lip (29%) and lateral aspects of the face (19%). The UCLP group also showed increases in size above the tip of the nose (25%) and laterally to the columella directly below the nares (29%).
Conclusions – Following surgical repair of UCLP in patients previously treated with NAM, 3D facial morphology was virtually indistinguishable from the non-cleft mean. Clinically, the apparent improvement in the facial soft tissues may mask dysmorphic skeletal growth, and further studies are required to characterize the underlying bony changes associated with the soft tissue changes reported here.

Key words: cleft; morphology; morphometrics; pre-surgical orthopedics

Introduction

Successful management of cleft lip and palate requires special attention to the soft tissues of the lip and nose, as well as the hard tissues of the maxilla, including dental abnormalities (1). It is thought that good alignment of the alveolar segments using non-surgical techniques can provide a foundation upon which the results of lip and primary nasal surgery can be built. For example, when using nasoalveolar molding (NAM) it is suggested that primary surgical repair of the nose and lip heals under minimal tension, reducing scar formation and improving the esthetic results (2–4). Thus, NAM is used to reshape or remodel the nasal cartilages and mold or remodel the maxillary arch before cleft lip repair and primary rhinoplasty. It has also been suggested that NAM provides esthetic benefits in terms of nasal tip and alar symmetry, and functional benefits in terms of improved dental arch form. Grayson and Cutting (5) reviewed the role of NAM in the primary correction of the nose, lip and alveolus in infants with bilateral and unilateral cleft lip and palate (UCLP). In addition, Santiago *et al.* (6) reported a 60% reduction in the need of secondary alveolar bone grafts in patients where pre-surgical NAM and a primary gingivoperiosteoplasty were combined as part of the treatment protocol. Later, Pfeifer *et al.* (7) suggested that the combination of NAM and gingivoperiosteoplasty is more cost-effective when compared with treatments involving secondary alveolar bone grafts.

Despite the claimed benefits of management using NAM, the clinical techniques employed do not follow a standard protocol. For example, Mitsuyoshi *et al.* (8) reported the use of a nasal stent constructed from cobalt–chrome wire, which is thought to enhance manual control of the forces and direction of the stent by the operator. Another modification that allows easier adjustment is the use of an orthodontic wire that projects from the palatal plate. It ends with an acrylic bulb, which is positioned inside the nares underneath the alar cartilage and acts as a nasal stent (9). Yet another approach includes a plate that utilizes the functional movements of the facial muscles to guide the major segment into a more normal position. The NAM is undertaken after correction of the alveolar position, and it is thought that this technique helps to improve alveolar position, nasal septum alignment, nasal symmetry, and nasal tip projection before lip repair (10). Thus, despite variations in clinical techniques, considerable success is claimed when deploying NAM.

To assess the efficacy of NAM, facial photographs were taken in one study after cheiloplasty and at age 1 year (11). Direct measurements were made on the photographs and when compared with their pre-surgical values improvements were found. However, some relapse of nostril shape was reported although no control subjects were included in that study. In another study (12), photographs were taken before and after NAM, after cheiloplasty, and yearly thereafter. In that study, linear measurements were made directly on the photographs, which suggested that nasal asymmetry was improved with NAM. However, after primary cheiloplasty, nasal asymmetry relapsed although no control subjects were included in that study either. Therefore, the aim of this study is to investigate 3D facial morphology following surgical repair of UCLP in patients after NAM compared with an age-, sex- and ethically matched control group. The hypothesis to be tested is that there are differences in facial morphology in patients surgically treated for UCLP after NAM compared to non-cleft infants. Rejection of the hypothesis will highlight the efficacy of a treatment protocol that relies on NAM to provide a foundation for a successful outcome in the early post-operative stage before further post-natal craniofacial morphogenesis.

Materials and methods

After obtaining IRB approval and HIPPA clearance, 15 infants with complete left-sided UCLP were imaged using 3D stereophotogrammetry (3dMD; LLC, Atlanta, GA, USA) before surgical treatment at the Center for Craniofacial Disorders, San Juan, PR, USA. All infants were imaged seated on the lap of the parent/care-giver and the distance from the tip of the infant's nose to the central beam of the imaging unit was adjusted to 90 cm. A series of images was taken when the subject was in a relaxed or normal state, with the infant's attention being attracted by a bright-colored toy placed directly on the central beam of the imaging unit. Images of subjects that were distressed were not included in the analysis. After processing, images that were suitable were edited to remove extraneous data and reoriented in the frontal plane using the imaging software before the placement of any landmarks. All infants had previously successfully completed a course of NAM (4). At a mean age of 37 weeks, all infants underwent lip repair and primary rhinoplasty by the same surgeon (MY). After 4 weeks post-operative resolution, all infants were re-imaged. As 3D stereophotogrammetry is a non-invasive, non-ionizing technique, repeat images pose no additional risk to the subject. In addition, 10 age-, sex- and ethically matched, non-cleft, control infants were similarly imaged, after parental consent was obtained as above. A power calculation for control sample size was undertaken using software (tpsPower; 13) that relates k landmarks in d dimensions to provide an appropriate n sample size. Using the landmark data, tpsPower indicated that with $n = 10$ in the control sample and standard deviations derived from the mean Procrustean forms, the null hypothesis of no difference in the means would almost always be rejected. This result was computed using Goodall's F-test, which makes very strict assumptions (e.g. independent isotropic error at all landmarks). Therefore, a control sample size of $n = 10$ is sufficient for statistical analysis using geometric morphometrics deployed in this present study. Thus, for this study, two groups were constructed: left complete UCLP ($n = 15$) and control infants (CON, $n = 10$).

Using appropriate software, five facial landmarks (Fig. 1) were digitized by the same investigator (GDS) on two different occasions. Procrustes superimposition was used to create a mean set of landmarks, followed by warping of the individual objects to the Procrustes mean, using a spline interpolation. Next, the dense correspondence of the splined objects was computed (transforming all vertices of the objects into landmarks), and an inverse spline was employed to effectively warp the objects back to their original state. Finally, Procrustes superimposition of all of the surface vertices was undertaken once more to create a mean surface. For outcome assessment, principal components analysis (PCA) and finite-element scaling analysis (FESA) were utilized. Details of these computing and analytical procedures are available (4, 14, 15).

Results

Duplicate digitization on two occasions yielded similar results ($p > 0.05$). Thus, the digitization error was not considered to be significant and further geometric morphometric analyses were warranted.

The mean UCLP face is shown in Fig. 2, while the mean CON face is shown in Fig. 3. Using PCA, it was found that the two groups were incompletely separated, as shown in Fig. 4, which depicts the first two eigenvalues that account for 63% of the total shape difference between the two groups. Thus, these first two principal components indicated that the UCLP and the non-cleft control groups show similar distributions in the modal space ($p > 0.05$), even though the UCLP

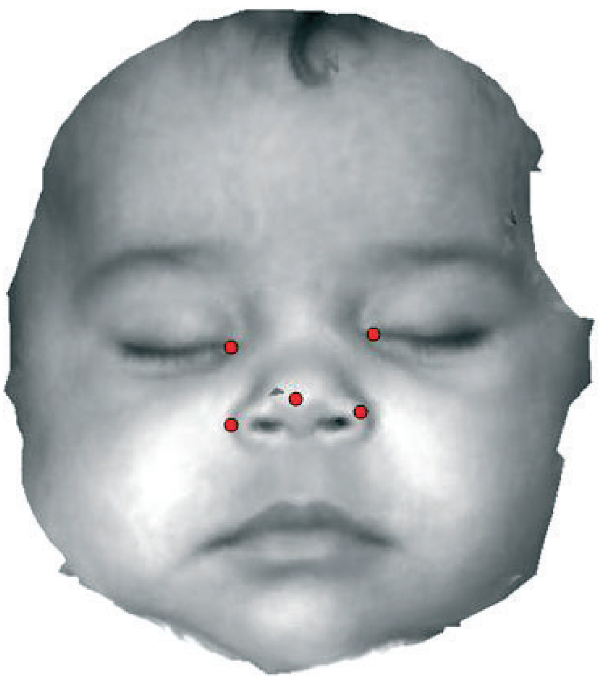

Fig. 1. The five facial landmarks that were digitized in the study were Pronasale, the right and left alar curvature, and the right and left medial canthus.

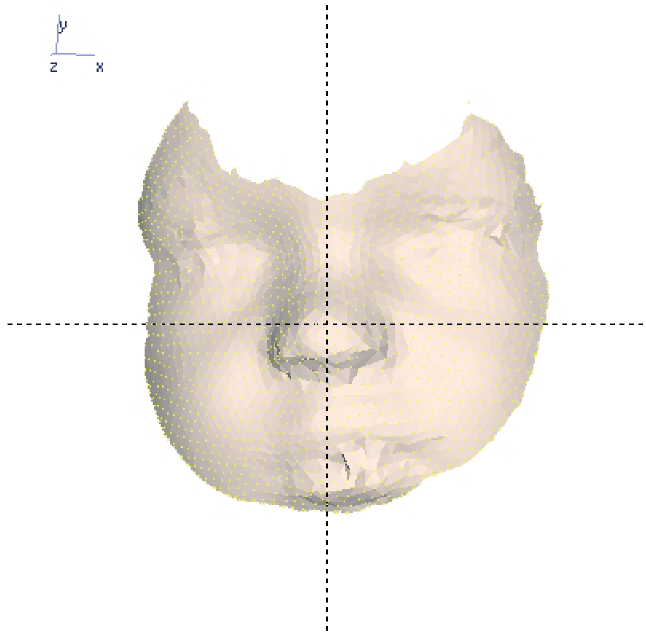

Fig. 2. The average three-dimensional (3D) model of the mean left-unilateral cleft lip and palate (UCLP) face reconstructed from the 15 patients imaged, i.e. the average face of the left-sided UCLP group of patients.

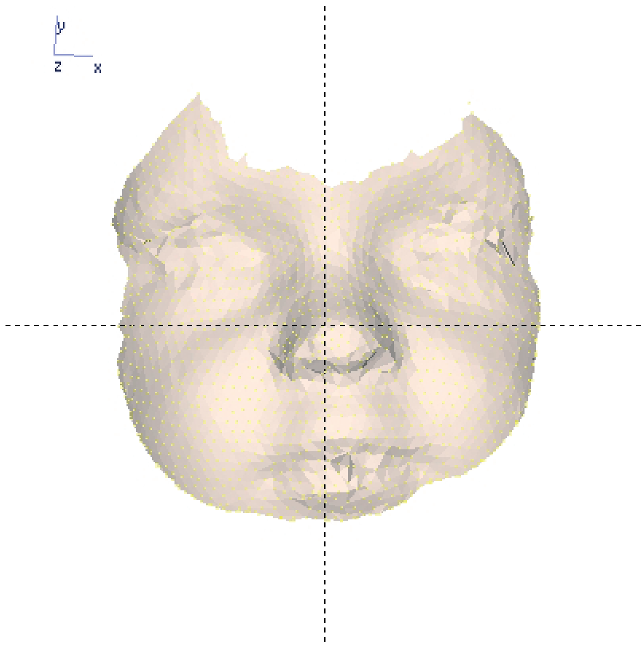

Fig. 3. The average 3D model of the mean control face reconstructed from the 10 subjects imaged, i.e. the average face of the control group of infants.

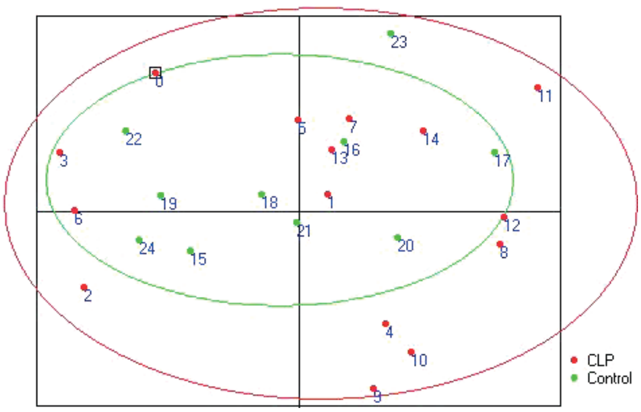

Fig. 4. Principal components analysis, showing the first two eigenvalues with the highest values, which indicate that using 63% of the shape information available, the two groups are incompletely separated.

group appears to exhibit a larger degree of shape variation, suggesting at least a minor degree of facial dimorphism.

To test the above notion, the mean UCLP 3D facial form (Fig. 2) was superimposed on the non-cleft control mean (Fig. 3). As shown in Fig. 5, the mean UCLP face appears to be smaller and less protrusive when compared with the control group, especially on the left side and in terms of nasal projection. Indeed, using pseudocolor FESA (Fig. 6), reductions in facial surface area were found in the UCLP group, involving the columella (29%), labial tubercle (51%), lower lip (29%) and lateral aspects of the face (19%). However, the UCLP group also showed

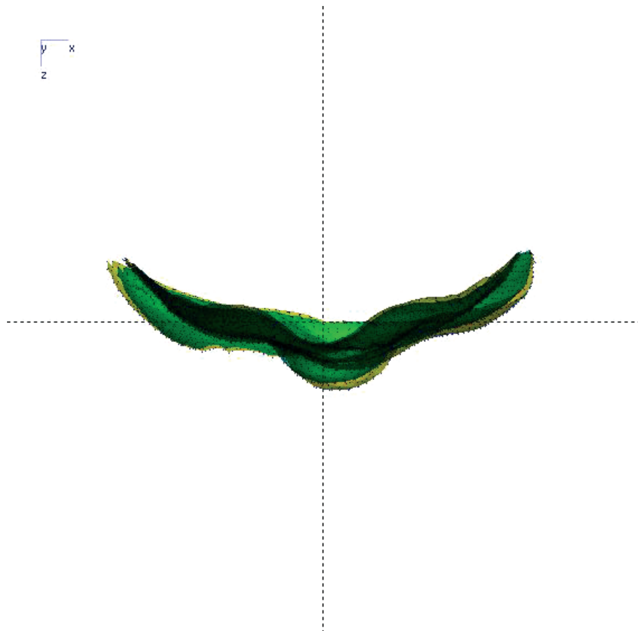

Fig. 5. The mean unilateral cleft lip and palate (UCLP) 3D facial form (green) superimposed on the non-cleft control mean (yellow) viewed from above. In this 'vertex' view, the mean UCLP face appears to be smaller and less protrusive when compared to the control group, especially on the left side, and in terms of nasal projection.

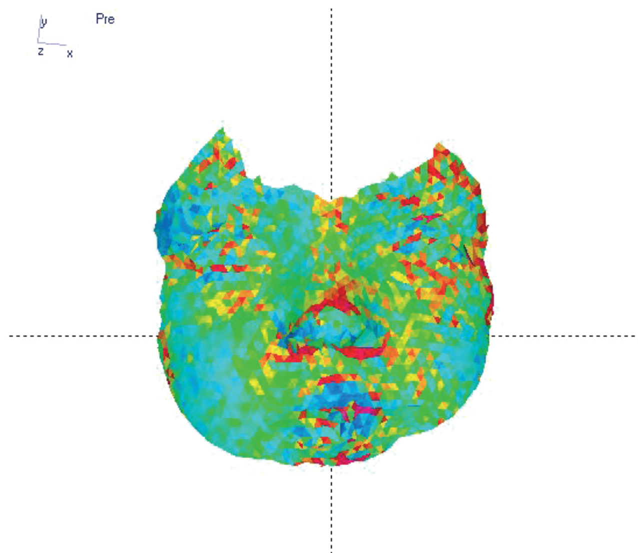

Fig. 6. Using pseudo-color finite-element scaling analysis to demonstrate changes, reductions in facial volume (colored blue) are found in the unilateral cleft lip and palate (UCLP) group, involving the columella (29%), labial tubercle (51%), lower lip (29%) and lateral aspects of the face (19%). However, the UCLP group also showed increases in size (colored red) above the tip of the nose (25%) and laterally to the columella directly below the nares (29%). A green color indicates no change in size.

increases in size above the tip of the nose (25%) and laterally to the columella directly below the nares (29%). Nevertheless, major differences in facial symmetry were not apparent between the two groups.

Discussion

The systematic errors of the imaging method employed in this study have been validated with a mean distance error of 0.04 mm and a RMS of 0.36 mm (16), and submillimeter accuracy (17). Random errors were addressed by duplicate digitization on two occasions, which yielded similar results ($p > 0.05$). Thus, the systematic and random digitization errors were considered in this study but found to be non-significant.

The NAM is undertaken after correction of the alveolar segments, and considerable success is claimed when deploying NAM. However, the use of NAM is limited to a few centers and the numbers of patients for a study of this nature are limited. In a previous study (4), improvements in nasal morphology following NAM before surgical correction were noted. Therefore, the present study was performed to study 3D facial morphology following surgical repair of UCLP in patients after NAM compared with a non-cleft, control group. Prasad *et al.* (18), using 3D dental casts, concluded that different regimens in the management of UCLP can significantly affect maxillary growth. Thus, the management of the patients in this study followed that of Cutting *et al.* (19), who also used a combined protocol of pre-surgical NAM with a one-stage lip, nose, and alveolus repair for bilateral clefts.

In a later study, Maull *et al.* (20) studied 3D nasal casts, concluding that NAM increases the symmetry of the nose. However, that study noted that asymmetry alone is not an adequate shape descriptor, and the control group was not age matched in that study *inter alia*. Therefore, in this present study an age-, sex- and ethically matched control group was included. It was found that following surgical repair of UCLP in patients previously treated with NAM, the overall UCLP facial morphology, including the nose, was improved to the extent that it was virtually indistinguishable from the non-cleft mean despite a wider range of shape variation. However, it was apparent that clinically the UCLP showed less nasal projection but this finding could simply reflect the smaller size of the UCLP patients, as it well documented that children with UCLP may be systematically smaller than their non-cleft counterparts.

Our findings are similar also to those of Wood *et al.* (21) who reported the effects of gingivoperiosteoplasty on midfacial growth following primary surgical repair. Using lateral cephalographs, Wood *et al.* (21) were unable to demonstrate impairment of maxillary growth in the patients treated with gingivoperiosteoplasty. However, it should be noted that our present results represent a 1-year time interval using 3D soft tissue data, while the previous study of Wood *et al.* (21) was carried out after 6 years in 2D. Nevertheless, the results of our current study suggest that the hypothesis that there is no difference in facial morphology in patients surgically treated for UCLP after NAM compared with non-cleft infants cannot be fully rejected.

While our study highlights the efficacy of a treatment protocol that relies on NAM, further post-natal craniofacial morphogenesis is reliant upon gene–environmental interactions, some of which presumably predispose to alterations in facial growth trajectories. Thus, the apparent reduction in facial volume clinically identifiable for the UCLP group noted here might be a predictor of latent dysmorphic skeletal growth patterns. It is not inconceivable that after successful initial management patients with surgically repaired UCLP may require further active therapy, such as functional orthodontic appliance treatments, if the initial advantages gained are not to be forfeited. Therefore, the apparent clinical differences in facial volume may be a harbinger of dysmorphic facial growth in children with UCLP, and further longitudinal studies are required to quantify underlying bony changes associated with the soft tissue changes reported here. In addition, future studies could include a post-surgical UCLP group that has been treated without NAM to determine the impact of NAM in patients with UCLP. If no differences between non-NAM and non-cleft patients are identified, then one would have to question the benefits of NAM reported in this present study.

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