

METHOD FOR OBTAINING A THREE-DIMENSIONAL IMAGE IN PATIENTS WITH CONGENITAL UNILATERAL CLEFT LIP AND PALATE

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INTRODUCTION.

Cleft lip is associated with a complex threedimensional, curved nasal deformity. Displacement of the nasal base elements results in a "tilted tripod", a deviated septum, and collapsed arcuate shapes that define the nose. Although primary correction is performed for initial cleft lip repair, recurrence is common, and 35 to 74 percent of children require a secondary revision. Given that current surgical practice is based on expert opinion and experience, an objective understanding of the initial deformity and the longitudinal changes that occur with treatment and growth is critical to improving medical care. In a separate report, we described an alternative approach to primary rhinoplasty and evaluated the associated three-dimensional nasal changes. An audit of the images revealed several surprising observations that challenge current thinking about the nasal deformity of

unilateral cleft lip. First, instead of a lateral displacement of the alar base cleft, it was not the alar base cleft, which was further from the facial midline. (Fig. 1.)

Second, instead of the alar base cleft moving medially with surgery, it was not the alar base cleft. Finally, instead of periodic re-growth of the alar base cleft over time, direct postoperative advancement was maintained. To formally investigate these observations, we undertook a detailed quantitative three-dimensional spatial analysis of the nasal base in order to assess the following:

1. Preoperative morphology of unilateral cleft lip nasal deformity.

2. Changes occurring in cleft lip repair and our approach to primary rhinoplasty.

3. Stability of surgical corrections over time

Fig. 1. Unilateral cleft lip, nasal deformity. Nasal alar

MATERIALS AND METHODS OF RESEARCH:

Patients who underwent permanent cleft lip repair and basic rhinoplasty from 2010 to 2013 were included in the study ($n = 102$). Surgical details are described in a separate report.8 Component reconstruction included approximation of the nasal threshold and lip as a whole, creation of a perineal muscle sling, primary septal repositioning, nasal floor closure and release, and reconstruction of the lateral lateral nasal walls. The approach seeks to create a firm foundation to address the root cause of the nasal deformity and does not involve opening up the tips. We accessed our center's database for 3D images (3dMD Cranial System; 3dMD, Atlanta, Ga.) Before, after surgery and at the end of follow-up. The images were acquired in the clinic by a professional imaging technologist at our center. To avoid the influence of other surgical interventions, we excluded images after secondary alveolar process transplantation or other procedures that may alter the shape of the nose. **Age-related controls.**

Three-dimensional images of children without clefts were obtained from Craniobank 15 (6 months; $n = 33$) and FaceBase (www.facebase.org) (Accession FB00000491; project U01DE020078) 16 (5 years; n = 40).

Image analysis.

Standard anthropometric landmarks 17 were identified using 3dMD Vultus (Fig. 2). Images were normalized along standard axes using the interendocanthion (enen) line to define the x-axis (medial-lateral) and the midpoint of this line to define the midline of the face. The images were rotated to the horizon defined by Tanikawa so that the y (craniocaudal) and z (anteroposterior) axes could be determined: the images were mounted with a 7.5-degree rotation downward from the subnasale-tragal axis (Fig. 2). Images of patients with right-sided clefts were reflected so that all images had clefts on the left side. Landmarks and changes with processing and time were evaluated along each axis and, if applicable, relative to the midline. The landmark positions on the cleft-free side were reflected across the defined facial midline to measure the relative displacement of the respective cleft-side landmarks and provide a symmetry analysis in a matched pair.

Fig. 2. Analysis of the three-dimensional image. (Left) Interendocanthion line is defined by the X-axis and the facial midline. Standard anthropometric landmarks included the following: endocanthion (en), pronasal (prn), alare (al), subalare (sbal), subnasale (sn), and chelion (ch). Images of subjects with clefts were normalized so that the cleft-free side (N) was on the right side of the face and the cleft side (C) was on the left side of the face. (Right) Images were normalized to a rotated Camper's plane [tragion (tr) to subnasal, -7.5 degrees] to determine the horizontal and y-axis and z-axis

RESULTS:

Figure 3. Subjects and demographics. Analysis of 289 images involving 87 subjects with cleft and 73 age-matched controls. 3D, three-dimensional.

Preoperative: Alar base displacement The approximate positions along the x and y axes are shown in Figure 4. Neither side of the nose was

normal when the subjects were compared with themselves and with the control group. The noncleavage sublareolar was lateral to the midline than the cleavage sublareolar; the cleavage sublareolar was further backward relative to the non-cleavage side. When the non-split sublareolar was reflected along the facial midline, the cleft sublareolar was 2.3 mm medial

(closer to the midline) and 1.5 mm below the reflected point. The asymmetry of the weakly expressed landmarks gradually increased for more severe cleft types with increasing transverse and superior displacement of the non-cleft sublare (Table 1).

sn, subnasal; sbal, subalar.

 $*$ Significant difference between the landmarks (p <0.02).

Fig. 4. Preoperative plot of landmarks along the x- and y-axes. The landmarks of the middle cleft subject are indicated by a red outline with the position of the nondurable sublare reflected along the midline marked in solid red (sbalN′). The landmarks of the middle-aged control subject correspond to the black outline, and the reflected sublare marked in solid black. The subnasale (sn) and non-split subalare (sbalN) were displaced superiorly and laterally, whereas the position of the split subalare (sbalC) was further medial and corresponded to that of the control subjects. en, endocanthion; prn, pronasal alare; al, alare; sbal, subalare; C, cleft side; N, week side

Preoperative: subnasal displacement leads to deformity

Among the evaluated landmarks, the single greatest displacement, compared with controls, was subnasal (Table 1). The mean X-axis displacement was 4.6 mm for all subjects (2.7 mm for incomplete and 6.2 mm for complete). We found that an increase in subnasal deviation was associated with an increase in cleft type severity and landmark displacement. Figure 6 illustrates the nature of the strain that occurred when the distance from the subnase to the midline was used as the dependent variable in the regression model. On the cleft side, there was no change along the x-axis, but there was progressively greater retro-positioning along the z-axis. On the cleft-free side, there was a lateral sublateral shift, but the expansion was proportionally less than the subnasal shift. As a result, the progressive dilation of the slit side of the nostril was associated with a progressive narrowing of the nostril without the cleft. In addition, we found that increased subnasal deviation from the midline was also associated with progressive widening of the intercanal distance. All associations were statistically significant $(p < 0.02)$. When the Bonferroni correction was applied, all associations remained significant except for intercanal dilation.

Changes with Surgery.

The greatest changes in magnitude were in the movement of the subnasal and week alar base positions (Figures 7 and 8). Both shifted medially. There was little medial movement of the cleft at baseline along the x-axis; however, there was significant anterior movement along the z-axis (Figures 7 and 9). The alar base and subnasal correction approached the correction of normal control on all axes. The cleft and nasal base positions were within 1 mm postoperatively when the landmarks were reflected along the midline (Table 1).

Stability of correction.

The symmetry of the alar base correction and midline subnasal position was maintained for 5 years, and was within 2 mm. Overcorrection of alar base retrusion was also stable (Fig. 9), with a sustained relative bulge of 1 to 2 mm. Compared with the control group, subjects with clefts had symmetrical enlargement of the alar base and loss of nasal projection. However, the symmetry of the alar base and the proximity of the subnasal to the midline were similar (Fig. 8).

Changes and stability for each type of cleft

The nasal base displacement gradually increased to increase the severity of the cleft.The reference positions were normalized after surgery regardless of cleft type. The variance of relative nasal base positions was within the same range as age-matched controls. Alar base retroversion was normalized for all cleft types and remained stable at age 5 years until secondary alveolar bone grafting.

DISCUSSION

Correction of nasal deformity of unilateral cleft lip remains a surgical problem without an optimal solution. An objective understanding of the primary deformity, changes in treatment, and changes with time and growth are key to implementing more ideal algorithms. The results of this study were unexpected and changed our perception of the primary deformity and reconstruction approaches (Table 2).

Preoperative nasal deformity

Cleft lip nasal deformity is classically described as a deformity arising from lateral and inferior displacement of the alarticular base on the cleft 9-14 side, with the cleft-free side considered normal. The conclusions of our study are not the same. Neither side is normal, and instead of an alar base cleft, it is not an alar base cleft that is laterally displaced relative to the facial midline (Figures 1). Based on our analysis of the threedimensional images, subnasal displacement is the single largest anomaly and correlates with progressive nasofacial deformity, which includes posterior displacement of the alar cleft base, lateral displacement of the non-alar cleft base, narrowing of the nostril width and widening of the intercanal

distance. In addition, contrary to previous reports that the septum was dislocated on the week side of the anterior nasal spine, we found that the caudal septum was always closely associated with the anterior nasal spine. Together, these findings suggest that the anterior nasal spine or caudal septum controls nasal deformity. We hypothesize that disruption of the horizontal support of the upper jaw leads to a "split" that rises along the bony skeleton. Mechanical forces from the upward thrust of the tongue and mandible, combined with unrelated growth of the premandibular bone, on the one hand, lead to projection and lateral torsion of the premandibular bone. The cleft of the alar base remains behind and the endowed alar base moves forward and sideways. This could explain the progressive narrowing of the nostril and the widening of the intercanal distance with a large offset of the subnasal from the midline. It also explains why the deformity of the palate and cecum visibly coincides with the shape of the tongue. The results of this study complement current models of cleft lip nasal deformity. Our observations are consistent with Latham's cadaveric studies in which the anterior nasal spine is the result of calcification of the septopremandibular ligament when there is fusion of the premandibular and lateral maxillary processes.

They confirm the displacement of the basic elements of the tilted tripod model1 and provide some insight into the patterns of this displacement. Fisher and Mann suggested that the nose consists of horizontal and oblique arch forms, which collapse when the base elements are displaced Fisher et al. later studied CT scans in 12 children with complete unilateral cleft lip and palate and described base element displacement

similar to what we found. Our study is different in that we studied 87 subjects who covered the spectrum of unilateral cleft lip and had a control group for comparison. We found progression of the deformity, which appeared to follow lateral deviation of the subnasal and "cleavage" of the facial skeleton. Given that expansion of the maxillary arch was found with a greater degree of cleft and our finding that the intercanal distance increased with deviation from the subnasal, the progressive "cleavage" of the facial skeleton is plausible and consistent with the concept of Latam pre-maxillary growth. The ascending deformity is also consistent with previous findings of upper lateral cartilage asymmetry, upper and lower cartilage dissociation, nasal bone deviation, and skull base asymmetry. Although reports of hypertelorism associated with oral cleft were inconsistent and contradictory,we found a clear progressive extension when subnasal deviation was used as a sign of cleft deformity across the spectrum of unilateral cleft lip presentation. Our findings and the concept of ascending facial skeletal deformity provide a common thread that can link previous models and observations concerning unilateral cleft deformity. **Surgical Modifications.**

We previously understood that repairing a cleft alar base involves release and medial, upper, and anterior positioning to fit the side without the cleft. Others have described the same thing. This would make intrinsic sense given the muscle release, wide dissection over the anterior maxilla, and complete release along the pear-shaped foramen, which all occur on the cleft side. In contrast to these expected changes, we found that it was not the alar base cleft that moved medially, whereas the alar base cleft moved only forward (and did not change its horizontal position) (Fig. 7). These movements were surprising, given that the non-cleft alar base cleft is outside the surgical site of dissection, and no suture is performed for the auricular alarum. Correction of the caudal septal position may explain the subnasal to midline movement. Further medial movement of the unexpanded alar base is probably due to the introduction of the lateral lip musculature into the dead space below the columella, thereby affecting the tension of the alar base together. Although the changes were not as expected, the surgical approach used in this study corrected the nasal base elements so that they were symmetrical and similar to those of control subjects

Figure 7. Example three-dimensional image of the subject before and after cleft lip repair with an underlying primary rhinoplasty. The cleft alar base is located posteriorly and moves forward. The unexpanded alar base and columella are displaced laterally and shifted medially.

Stability of correction Recurrent or permanent cleft lip nasal deformity is due to recurrent or residual alar base rejection. Bone

anatomy studies using computed tomography show persistent anterior nasal deviation and cleft sternal process after cleft lip repair and after 10 years of age. . Thus, it is generally believed that definitive nasal correction requires correction of the bony base by secondary alveolar bone grafting.We found that threedimensional alar base symmetry can be achieved immediately after primary reconstruction and that symmetry of nasal base correction persists with time and growth in all planes, including the z axis (anteroposterior).This was true regardless of cleft type or bone defect width. The follow-up period was 5 years and was before alveolar process transplantation. Based on these results, a stable anteroposterior correction of the alar base can be achieved without regard to the bony base. Although we found that the symmetry of the nasal base correction persisted with time and growth, we found that both alar bases tended to expand with reduced pronasal projection compared to control subjects of the corresponding age. There may be several reasons for the differences between the cleft and control group. First, longitudinal three-dimensional images of control subjects were not available, and we used two sets of control images that included different racial demographics for comparison. Images from Craniobank (used to compare cleft subjects immediately before and after surgery)

included a large number of African Americans, whereas images from FaceBase (used to compare cleft subjects at 5 years of age) consisted of subjects of European Caucasian descent. Although facial symmetry may be expected according to racial variation, nasal width and facial projection may differ, which explains some of the differences. The second may be that there is a gradual stretching of the tissues after reconstruction, so that the distance between the lumen increases and the tip of the nose loses projection. The third may be that there is some degree of growth reduction in subjects with cleft relative to control subjects. Hyperplasia may be characteristic of oral cleft. It is also generally accepted that scarification from surgery has a cumulative effect on facial growth. This effect has been found in both cleft lip and cleft palate reconstruction. We cannot determine whether any additional change in growth is a result of the approach we used to correct the nose. Previous studies have not demonstrated any greater growth disruption due to primary septoplasty or fundus closure relative to children who undergo surgery without these adjectival procedures. Future studies to formally examine facial growth patterns are needed

Table 2. Paradigms

Fig. 8 Unilateral cleft deformity (same patient as Fig. 1). The cleft of the alar base is retracted, but normal in the horizontal position. The nonnatal alar base and columella are deviated to the side. Correction requires moving each component along the appropriate vectors (red arrows). Preoperative morphology can give an idea of the "forces" at play. The vomer / septum is visible through the cleft, but collapsed so that the most caudal edge deviates from the midline. A "fold" / section of vertical collapse is visible, and the combined palatal and vomer-septal surfaces visibly take the shape of the tongue. Loss of horizontal integrity of the maxilla can make the facial skeleton prone to collapse and expansion of the vertical supports and an upward "split." Irritated premandibular growth on one side causes anterior rotation.

Approaches to the correction of cleft lip nasal deformity

Given that the most obvious and recognizable feature of cleft lip nasal deformity is the asymmetrical dissociation and collapse of the lower lateral cartilages, correction of the nasal tip has become a focal point of primary rhinoplasty. Although significant improvement occurs when cartilage is inserted into the skin, permanent or permanent disfigurement is common. This may be due to an untreated bone base or "base." Regardless of these perceptions, the results of this study change some of our fundamental assumptions about primary unilateral cleft lip nasal deformity and the changes we must make in surgical correction. The cleft alarm does not shift laterally; rather, the lower nose is deflected and deviated from the midline. If we want to correct this primary deformity, we need to make the appropriate changes.

CONCLUSIONS

Caudal septal deviation, as measured by subnasal offset from the midline, results in a unilateral cleft lip nasal deformity. The alarial base with a non-zero displacement is lateral, whereas the alarial base of the cleft is posterior. The nostril nostril narrows and the intercanal distance widens with greater deformity. The underlying primary rhinoplasty involves significant medial movement of the fundus and columella with anterior movement of the alar base cleft. In this study, the approach corrected nasal base asymmetry in all axes (x, y, and z). Postoperative nasal base symmetry was similar to that of control subjects and persisted with time and growth.

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