Three-Dimensional Analysis of Craniofacial Structures of Individuals With Nonsyndromic Unilateral Complete Cleft Lip and Palate

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Abstract: Cleft lip and palate (CLP) is one of the most common congenital deformities. Primary surgeries at an early age result in scar formation, which may impede the growth of craniofacial structures of the maxilla. Orthodontist's role in the management of individuals with CLP is important and starts from the time of birth. The knowledge of craniofacial structures in individuals with a cleft is essential for treatment planning. The purpose of this study was to analyze and compare craniofacial structures of cleft and noncleft side of individuals with non-syndromic unilateral complete cleft lip and palate (NSUCCLP) using cone-beam computed tomography (CBCT). CBCT scans of individuals with NSUCCLP (n = 42) were retrieved from the databases of two cleft centers, which followed the same protocols for timing and type of primary surgeries and secondary alveolar bone grafting (SABG). DICOM files of CBCT scans were integrated into Dolphin 3D software, and analysis was carried out in multiplanar views. The craniofacial structures of individuals with NSUCCLP were analyzed using fourteen parameters. Measurements were also recorded between the cleft and noncleft sides for comparison. The volume of the maxilla was generated by isolating it from adjacent structures on a 3D reconstructed model. MAWC, MAWPM1, MAWPM2, MAWM1, and MV of the cleft side was less than noncleft side ($P < 0.05$). MHP @ N Aper is less on the noncleft side ($P < 0.05$). There is an asymmetry of structures around the dentoalveolar and nasal region; however, asymmetries were not affected at deeper structures of the craniofacial region of individuals with NSUCCLP.

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Cleft lip and palate (CLP) is one of the most common congenital deformities which results from the failure of fusion of the maxillary and palatine processes.[1,2] Cleft can involve both lip and palate or either lip or palate. Based on the type and site of involvement, it can be classified as complete or incomplete, unilateral or bilateral. Clefts can be syndromic or non-syndromic. A cleft is called syndromic if the patient has malformation, which involves more than one developmental field. A cleft is said to be non-syndromic, if there is a malformation that is the resultant of a single initiating event, involving one developmental field.[3] 70% of individuals with CLP and 50% of individuals with cleft palate only (CPO) are non-syndromic in nature.[2] The causes of non-syndromic unilateral complete cleft lip and palate (NSUCCLP) remain largely unknown. Clefts have a complex etiology with both genetics and environment playing an important role. Risk factors such as folic acid deficiency, maternal age and smoking have been linked to the development of clefts.[4–6]

The management of CLP involves a fully integrated dedicated cleft team comprising of pediatricians, cleft surgeons, orthodontists, geneticists, social workers, ENT, speech therapists, prosthodontists, psychologists and oral hygienists.[7] The orthodontist plays a pivotal role in the management of individuals with CLP and orthodontic intervention can be categorized into several phases.[7–9] Keeping long-term treatment needs in mind, orthodontists should have sound knowledge of craniofacial structures in patients with UCCLP.

The surgical repair of cleft lip and palate during infancy and early childhood improves facial appearance and functional development, and it can cause maxillary deficiency.[10,11] The assessment of craniofacial structures in patients with UCCLP has been the subject of research and evaluation for the past many years. Several researchers reported long term studies of craniofacial structures in individuals with CLP using 2D imaging tools.[12–19] 2D imaging tools fall short in assessing craniofacial structures in its entirety due to limitations, which include superimposition and magnification.[20–21] The deeper structures cannot be studied separately for the cleft and noncleft side. With the advent of 3D imaging tools, it is now possible to obtain detailed and accurate views of a structure at any level.[20,21]

Cleft lip and palate is a 3 Dimensional (3D) facial deformity, and 3D imaging would provide a better insight into the anatomical structures. Computed tomography (CT) and cone-beam computed tomography (CBCT) are the commonly used 3D imaging tools for evaluation of the craniofacial structures. Some studies have analyzed the effect of craniofacial structures using CT in individuals with cleft.[22–24] CBCT has good applicability in individuals with CLP as compared to CT because of lesser radiation and low cost.[25] Published literature also reported that CBCT imaging provides a good diagnostic tool for quantifying and analyzing surface and deep craniofacial structures in individuals with CLP.[26,27] With the availability of third-party software, Digital Imaging and Communications in Medicine (DICOM) files can be integrated into the software, making it possible to analyze cleft and noncleft sides separately.

CBCT images have been found supportive in better understanding of diagnosis and treatment planning in an individual with CLP. Although 3D imaging has been a useful tool to study the deformity, its main drawback is increased radiation dose. Whenever CBCT has to be prescribed after the risk-benefit assessment, it is recommended to follow As Low As Reasonably Achievable (ALARA) principle.[28] SEDENTEXCT justifies CBCT prescription in patients with CLP over MSCT.[29] The purpose of this study was to analyze the superficial and deeper craniofacial structures in individuals with NSUCCLP using CBCT.

METHODOLOGY

This retrospective cross-sectional study was carried out after approval from the University Ethics Committee. Permission was obtained from the two craniofacial centers to use CBCT scans from their repositories. In order to obtain a homogeneous sample, specific criteria were used while selecting samples such as the age of the patient, timing and type of primary surgeries. A single operator examined the CBCT scans of individuals with NSUCCLP from the database of the two cleft centers which were obtained for treatment planning.

Inclusion Criteria for the Selection of the Sample Were as Follows

1. Individuals with NSUCCLP (Veau's group III type with negative overjet less than 5 mm)
2. All scans had a full field of view (FOV) which recorded the craniofacial structures completely
3. Availability of detailed records of treatment
4. Growing children between 10 and 15 years
5. History of (H/o) type and timing of primary surgeries of cleft lip and palate
6. H/o timing of secondary alveolar bone grafting (SABG).

The Surgical Protocol Followed for Primary Surgeries and Secondary Alveolar Bone Grafting

A proforma was completed from the existing treatment records of each patient, recording the age, site of the cleft, age of surgical repair of lip and palate and SABG. Both the centers followed the same protocols for surgical closure of lip and palate and SABG (Supplementary Digital Content, Table 1, http://links.lww.com/SCS/B732).

1. *Primary lip closure using morphofunctional cleft lip repair. (Afroze surgical incision technique, minimum age of 4 months and minimum weight of 5.5 Kg)[30]
2. *Closure of the palate was carried out using criteria for morphofunctional cleft palate repair. (One stage palatoplasty with the Bardach two flap technique with optimal muscle dissection, minimum age of 11 to 13 months and minimum weight of 8.5 kg)[31]
3. *SABG was carried out using autogenous iliac crest at 7 to 9 years before the eruption of the canine

The sample consisted of 42 individuals with NSUCCLP [males (n = 26); females (n = 16)] with a mean age of 12.12 ± 1.4 years, [cleft on right (n = 13), and cleft on left (n = 29)] (Supplementary Digital Content, Table 1, http://links.lww.com/SCS/B732). There is no significant difference in the mean ages of male or female groups or mean ages of cleft affected on right or cleft affected on left groups (Supplementary Digital Content, Table 1, http://links.lww.com/SCS/B732).

It was confirmed that all the participants and parents whose CBCT scans were used in this study were earlier provided with Participants Information Sheet (PIS) and consent/assent was obtained to utilize their data for research. Data which was blinded and anonymized was collected by one of the investigators.

Image Acquisition

The CBCT had been performed using 2 different machines with the following imaging conditions:

1. *Promax 3D scanner Mid with Proface (Planmeca, Helsinki, Finland)*: 20" × 17" FOV; 0.3 mm^3 (isotropic) voxel size; 90 kVp tube voltage; 14 mA tube current; 12 s scanning time: 12 s exposure time.
2. *NewTom 5G, QR, Verona, Italy*: 16' × 16' FOV; voxel size 0.3 mm^3 (isotropic); 90 kVp tube voltage, 10 mA; 10.8 s scanning time; 10.9 s exposure time.

The CBCT images were transformed to DICOM files and were imported into Dolphin 3D software (version 11.9, Build 24, Dolphin Imaging and Management Solutions, Chatsworth, CA) whose accuracy, reliability, and reproducibility in making linear measurements has been previously studied.[32,33] Images were analyzed in sagittal, axial, coronal slices, and the 3D reconstructed views. The craniofacial structures were analyzed using fourteen parameters. The parameters (Fig. 1 and Fig. 2) used to study craniofacial structures were adopted from previous studies, which were described in Supplementary Digital Content, Table 2, http://links.lww.com/SCS/B732.[24,34–35] Each parameter was standardized before analysis. The deeper structure analyzed in the study were FW, Cd W, Mx D, MHP @ N Aper and MHP @ Alv Cr. (Fig. 1F to Fig. 1I, Supplementary Digital Content, Table 2, http://links.lww.com/SCS/B732).

ML, PalThAnt, PalThPost, MWC, MWPM1, MWPM2, MWM1, FW and CW were analyzed whereas MAWC, MAWPM1, MAWPM2, MAWM1, MxD, MHP @ N Aper, MHP @ Alv Cr, NW, FW, CdW and MV were compared between cleft and noncleft side.

Statistical Analysis

All statistical analyses were performed using the SPSS software package (SPSS Statistics for Windows, V23.0; IBM, Armonk, NY). All the measurements were repeated twice with an interval of a month, and intra operator reliability was assessed with intraclass correlation coefficients (ICC). Shapiro-Wilk and Kolmogorov-Smirnov test were performed to test the normality of the data distribution. As data was following normal distribution after normality tests ($P > 0.05$), parametric tests were applied. The descriptive statistics (Mean and SD) were used to assess craniofacial structures (Supplementary Digital Content, Table 3, http://links.lww.com/SCS/B732). Paired *t* test was used to compare the measurements carried out on structures of cleft and noncleft side (Supplementary Digital Content, Table 4, http://links.lww.com/SCS/B732). Independent *t* test was applied to assess the difference in the measurements between male and female groups or

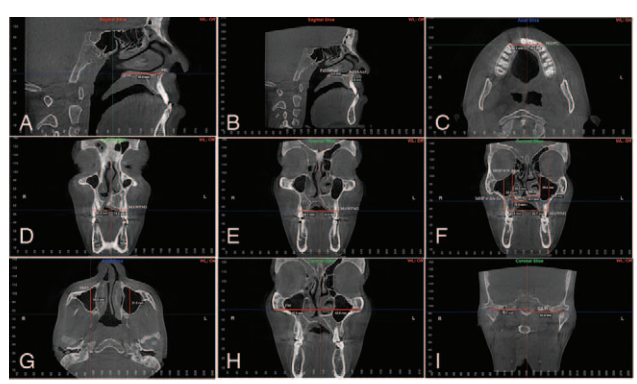

FIGURE 1. A: Maxillary length. B: Palatal thickness anterior and palatal thickness posterior. C: Maxillary arch width canine. D: Maxillary arch width premolar 1. E: Maxillary arch width premolar 2. F: Maxillary arch width molar 1, maxillary height posterior at alveolar crest, nasal width, maxillary height posterior at nasal aperture. G: Maxillary depth. H: facial width. I: Condylar width.

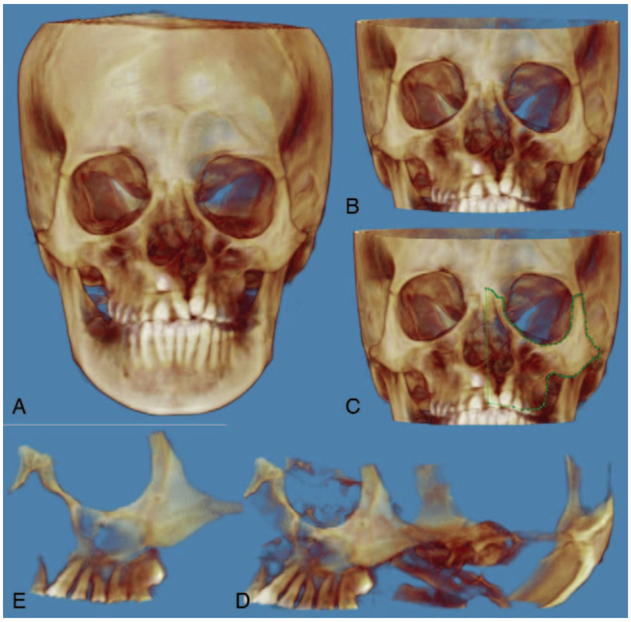

FIGURE 2. Maxillary volume; A. Complete 3D reconstructed model, B. Segmented upper half of the model, C. Outline of maxillary boundary, D. Segmentation of maxillary boundary, E. Isolated maxilla.

to analyze the difference in the measurements between UCCLP(R) and UCCLP(L) (Supplementary Digital Content, Table 5, http://links.lww.com/SCS/B732). $P < 0.05$ was considered to be statistically significant.

RESULTS

All craniofacial structure measurements showed excellent intra rater correlation (ICC = 0.91 to 1.00). Descriptive statistics of parameters were shown in (Supplementary Digital Content, Table 3, http://links.lww.com/SCS/B732). Paired t-test showed that MAWC, MAWPM1, MAWPM2, MAWM1and MV of the cleft side was less as compared to the noncleft side ($P < 0.05$) (Supplementary Digital Content, Table 5, http://links.lww.com/SCS/B732) M4HP @ N Aper is less on the noncleft side than on the cleft side. Male and female groups comparison showed no significant difference between the measurements except for CdW ($P > 0.05$) (Supplementary Digital Content, Table 5, http://links.lww.com/SCS/B732). Cleft affected on the right, or left side showed no significant variations in measurements for craniofacial structures except for MAWC, MAWPM1 and Cd W (Supplementary Digital Content, Table 5, http://links.lww.com/SCS/B732).

DISCUSSION

Primary surgeries of the palate may hamper the growth of the maxilla and the associated structures in individuals with CLP, leading to facial asymmetry in all the 3 planes. The asymmetry is more evident in individuals with UCCLP and requires correction. Assessment of craniofacial structures in such individuals is essential for treatment planning. Lateral Cephalograms (Lateral Ceph), posteroanterior radiographs (PA), orthopantomograms (OPG), 2D and 3D photographs, optical surface scans, CT, and CBCT are used to assess craniofacial structures in individuals with cleft.[36–52] Analysis in all 3 planes was possible after the introduction of 3D tools and CBCT is an evolved 3D tool for the assessment of craniofacial structures.

Cone-beam computed tomography scans of individuals with NSUCCLP (n = 42), before the commencement of orthodontic therapy of mean age 12.12 ± 1.4 years were analyzed in the study (Supplementary Digital Content, Table 1, http://links.lww.com/SCS/B732). This is the age when comprehensive orthodontic treatment begins for individuals with CLP. The sample showed there was no significant difference between the mean ages of boys and girls or cleft affected on the right or left (Supplementary Digital Content, Table 1, http://links.lww.com/SCS/B732). This study evaluated the craniofacial structures of the superficial and deep region to assess asymmetries in individuals with NSUCCLP. The present study showed that the asymmetries were found in the dentoalveolar and the nasolabial region (Supplementary Digital Content, Table 4, http://links.lww.com/SCS/B732).

Several studies in the literature have also reported that asymmetries were confined to the nasolabial and dentoalveolar region of individuals with UCCLP.[22,23,37,43–46,48,49,52]

Choi et al, compared the asymmetries of the cleft and noncleft sides in all the three planes using grid analysis in individuals with UCCLP.[37] The conclusions drawn by them were similar to the present study. However, their results did not reveal significant asymmetry in the deeper regions of the maxillary complex. Yang et al compared facial asymmetry of individuals with UCCLP and the control group using asymmetry index and noted significant differences around the cleft and the nasal chamber of the affected and the nonaffected sides.[46]

Starbuck et al compared the craniofacial structures of cleft and control group using morphometric analysis and reported asymmetry of the nasal regions of the midface. The morphometric differences of the upper and lower facial skeletons were found to be less affected.[48] Furthermore, Ahmad and Starbuck evaluated facial directional asymmetry (DA) using Euclidean Distance Matrix Analysis (EDMA) of the same sample. They reported that DA was greater in persons with UCLP group. Regions derived from the maxillary and nasal prominences demonstrated the most DA.[49]

Suri et al and Li et al analyzed craniofacial characteristics of individuals with UCCLP to compare the asymmetry of the cleft and non-cleft sides using spiral CT. They also reported that asymmetries were in the dentoalveolar and the nasal region.[22,23]

Kuijpers et al carried out a stereophotogrammetric study in individuals with a unilateral cleft to evaluate the most asymmetric area of the face.[45] They found that in individuals with UCLP, the most affected region was the nose, followed by the lips, chin, and cheek. Asymmetry in CLP patients using stereophotogrammetric method had also been reported in by Ras et al with similar results.[43,44] Zemann et al reported asymmetries in the orbital, the nasal, and the maxillary region.[52]

Due to the variation in methodology or imaging modality used (2D radiographs, CT, CBCT, optical surface scan, or 3D photogrammetry, etc.), or the parameters assessed, it was not possible to compare directly our results with results from other reported studies. The present study compared craniofacial structures of the cleft and noncleft sides of the same patient, whereas some of the reported studies compared cleft individuals with a matched control group. However, some of our parameters were compared with the parameters of 2 reported studies (Supplementary Digital Content, Table 6, http://links.lww.com/SCS/B732).[24,34]

Agarwal et al used CT to analyze the craniofacial structures of cleft and noncleft side in patients with UCLP and reported marked asymmetry between cleft and noncleft side.[25] Schneiderman et al used a set of parameters for a comprehensive analysis of the craniofacial structures using CBCT.[34] Though his study did not show a significant difference between the measurements of the two groups due to the small sample size, his study made an attempt to evaluate the symmetry between affected and non-affected sides.

Different investigators defined ML differently. In this study, we followed Burstone's definition of ML for the analysis, which is the distance from ANS to PNS.[53] Similar measurements from ANS and

PNS were referred to as palatal plane length (PPL) by Schneiderman et al. The ML of the present study was less than that reported by Schneiderman et al. The difference in measurements could be due to the variation in the race and age of selected participants. However, the values of ML reported by Agarwal et al (measured from maxillary tuberosity to the anterior contour of the maxilla) was similar to the values of this study. PalThAnt, PalThPost values of the present study were similar to that reported by a Schneiderman et al. Measurements for MAWC in the present study were similar to values reported by Schneiderman et al. and slightly less than values reported by Agarwal et al. All 3 studies recorded the same measurement for MH; however, values recorded for MHP @ Alv Cr in the present study was slightly less as compared to Schneiderman et al. MxD recorded in this study was similar to that recorded by Agarwal et al and less than the values recorded by Schneiderman et al.

Although the volume is an accurate predictor to determine deficiency, the analysis of volume was challenging in the past. Dolphin 3D software uses the increased bit-depth to improve its primary and secondary reconstructions, resulting in a cleaner and more defined volume. The segmentation feature of the Dolphin 3D software can be used to define the boundaries accurately to generate the volume of a 3D reconstructed model.[32]

The present study found significant asymmetries around the nasolabial and dentoalveolar region and the deeper structures less severely affected. This superficial midfacial asymmetry which influences the visual perception and the appearance of individuals can be corrected by protraction and expansion during growth and if necessary, with rhinoplasty at a later date. Although this study is cross-sectional, these parameters may be used to assess and analyze changes after therapy.

Limitation of this Study

Although this study aimed at a comprehensive assessment of craniofacial structures, it did not include an assessment of mandibular structures other than Condylar Width (Cd W). Control group was not used in the present study because of stringent rules and guidelines of CBCT prescription

Further Scope

Hemifacial cephalograms can be generated from CBCT using the cleft or noncleft side orientation to compare cephalometric parameters of each side separately.

CONCLUSION

This study analyzed and compared craniofacial structures in individuals with NSUCCLP using CBCT. The following conclusions are drawn from the present study.

1. MAWC, MAWPM1, MAWPM2, MAWM1, and MV was less on the cleft side than the noncleft side
2. MHP@ N Aper was more on the cleft side than the non-cleft side
3. Most of the asymmetries lie in the dentoalveolar and nasolabial region, and deeper structures were not affected.
4. Females recorded smaller measurements as compared to males; however, there was no significant difference in the measurement of the male and female groups except for CdW
5. Comparison of cleft affected on the right or left showed no significant difference in measurements except for MAWC, MAWPM1, and CdW.

REFERENCES

1. Cobourne MT. The complex genetics of cleft lip and palate. *Eur J Orthod* 2004;26:7–16
2. Mossey PA, Little J, Munger RG, et al. Cleft lip and palate. *Lancet* 2009;374:1773–1785
3. Prabhu S, Krishnapillai R, Jose M, et al. Etiopathogenesis of orofacial clefting revisited. *J Oral Maxillofac Pathol* 2012;16:228–232
4. Murray JC1. Gene/environment causes of cleft lip and/or palate. *Clin Genet* 2002;61:248–256
5. Honein MA1, Rasmussen SA, Reefhuis J, et al. Maternal smoking and environmental tobacco smoke exposure and the risk of orofacial clefts. *Epidemiology* 2007;18:226–233
6. Beaty TH, Ruczinski I, Murray JC, et al. Evidence for gene-environment interaction in a genome wide study of nonsyndromic cleft palate. *Genet Epidemiol* 2011;35:469–478
7. Kuijpers-Jagtman AM. The orthodontist, an essential partner in CLP treatment. *B-ENT* 2006;2 Suppl 4:57–62
8. Cash AC. Orthodontic treatment in the management of cleft lip and palate. *Front Oral Biol* 2012;16:111–123
9. Shetye PR. Orthodontic management of patients with cleft lip and palate. *APOS Trends Orthod* 2016;6:281–286
10. Tindlund RS, Rygh P, Bøe OE. Orthopedic protraction of the upper jaw in cleft lip and palate patients during the deciduous and mixed dentition periods in comparison with normal growth and development. *Cleft Palate Craniofac J* 1993;30:182–194
11. Liao YF1, Mars M. Long-term effects of palate repair on craniofacial morphology in patients with unilateral cleft lip and palate. *Cleft Palate Craniofac J* 2005;42:594–600
12. Aduss H. Craniofacial growth in complete unilateral cleft lip and palate. *Angle Orthod* 1971;41:202–213
13. Hayashi I, Sakuda M, Takimoto K, et al. Craniofacial growth in complete unilateral cleft lip and palate: a roentgeno-cephalometric study. *Cleft Palate J* 1976;13:215–237
14. Smahel Z, Mullerova Z. Craniofacial morphology in unilateral cleft lip and palate prior to palatoplasty. *Cleft Palate J* 1986;23:225–232
15. Smahel Z, Machovav P, Mullerova Z, et al. Growth and development of the face in unilateral cleft lip and palate during prepubertal and pubertal periods. *Acta Chirurgiae Plasticae* 1992;34:163–177
16. Smahel Z, Mullerova Z. Postpubertal growth and development of the face in unilateral cleft lip and palate as compared to the pubertal period: a longitudinal study. *J Craniofac Genet Dev Biol* 1996;16:182–192
17. Hoswell BB, Levant BA. Craniofacial growth in unilateral cleft lip and palate: skeletal growth from 8 to 18 years. *Cleft Palate J* 1988;25:114–121
18. Semb G. A study of facial growth in patients with unilateral cleft lip and palate treated by the Oslo CLP team. *Cleft Palate Craniofac J* 1991;28:1–21
19. Ross RB. Treatment variables affecting facial growth in complete unilateral cleft lip and palate. *Cleft Palate J* 1987;24:5–77
20. Quintero JC, Trosien A, Hatcher D, et al. Craniofacial imaging in orthodontics: historical perspective, current status, and future developments. *Angle Orthod* 1999;69:491–506
21. Erten O, Yilmaz BN. Three-dimensional imaging in orthodontics. *Turk J Orthod* 2018;31:86–94
22. Suri S, Utreja A, Khandelwal N, et al. Craniofacial computerized tomography analysis of the midface of patients with repaired complete unilateral cleft lip and palate. *Am J Orthod Dentofacial Orthop* 2008;134:418–429
23. Li H, Yang Y, Chen Y, et al. Three-dimensional reconstruction of maxillae using spiral computed tomography and its application in postoperative adult patients with unilateral complete cleft lip and palate. *J Oral Maxillofac Surg* 2011;69:e549–e557
24. Agarwal R, Parihar A, Mandhani PA, et al. Three-dimensional computed tomographic analysis of the maxilla in unilateral cleft lip and palate: implications for rhinoplasty. *J Craniofac Surg* 2012;23:1338–1342
25. Scarfe WC, Farman AG, Sukovic P. Clinical applications of cone-beam computed tomography in dental practice. *J Can Dent Assoc* 2006;72:75–80
26. Kuijpers MA, Chiu YT, Nada RM, et al. Three-dimensional imaging methods for quantitative analysis of facial soft tissues and skeletal morphology in patients with orofacial clefts: a systematic review. *PLoS One* 2014;9:e93442
27. Parveen S, Husain A, Mascarenhas R, et al. Clinical utility of cone beam computed tomography in patients with cleft lip palate: Current

perspectives and guidelines. *J Cleft Lip Palate Craniofac Anomal* 2018;5:74–87
28. Farman AG. ALARA still applies. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2005;100:395–397
29. European Commission. Cone Beam CT for Dental and Maxillofacial Radiology: Evidence-Based Guidelines. Luxembourg: SEDENTEXCT; 2012. http://www.sedentexct.eu/content/guidelines-cbct-dental-and-maxillofacial-radiology. Accessed June 10, 2020
30. Reddy GS, Reddy RR, Pagaria N, et al. Afroze incision for functional cheiloseptoplasty. *J Craniofac Surg* 2009;20 Suppl 2:1733–1736
31. Agrawal K. Cleft palate repair and variations. *Indian J Plast Surg* 2009;42 Suppl((Suppl)):S102–S109
32. Dolphin https://www.dolphinimaging.com/product/ThreeD. Accessed June 13, 2020
33. Fernandes TM, Adamczyk J, Poleti ML, et al. Comparison between 3D volumetric rendering and multiplanar slices on the reliability of linear measurements on CBCT images: an in vitro study. *J Appl Oral Sci* 2015;23:56–63
34. Schneiderman ED, Xu H. SalyerKE.Characterization of the maxillary complex in unilateral cleft lip and palate using cone-beam computed tomography: a preliminary study. *J Craniofac Surg* 2009;20 Suppl 2:1699–1710
35. Hwang S, Jeong S, Choi YJ, et al. Three-dimensional evaluation of dentofacial transverse widths of adults with various vertical facial patterns. *Am J Orthod Dentofacial Orthop* 2018;153:692–700
36. Laspos CP, Kyrkanides S, Tallents RH, et al. Mandibular and maxillary asymmetry in individuals with unilateral cleft lip and palate. *Cleft Palate Craniofac J* 1997;34:232–239
37. Choi YK, Park SB, Kim YI, et al. Three-dimensional evaluation of midfacial asymmetry in patients with nonsyndromic unilateral cleft lip and palate by cone-beam computed tomography. *Korean J Orthod* 2013;43:113–119
38. Mølsted K, Dahl E. Asymmetry of the maxilla in children with complete unilateral cleft lip and palate. *Cleft Palate J* 1990;27:184–192
39. Ishiguro K, Krogman WM, Mazaheri M, et al. A longitudinal study of morphological craniofacial patterns via P-A x-ray headfilms in cleft patients from birth to six years of age. *Cleft Palate J* 1976;13:104–126
40. Kyrkanides S, Klambani M, Subtelny JD. Cranial base and facial skeleton asymmetries in individuals with unilateral cleft lip and palate. *Cleft Palate Craniofac J* 2000;37:556–561
41. Kyrkanides S, Richter L. Mandibular asymmetry and antigonial notching in individuals with unilateral cleft lip and palate. *Cleft Palate Craniofac J* 2002;39:30–35
42. Keijser K, Nowinski D, Mani M. Photographic measurements partially correlate to nasal function and appearance among adult cleft patients. *Plast Reconstr Surg Glob Open* 2016;4:e720
43. Ras F, Habets LL, van Ginkel FC, et al. Three-dimensional evaluation of facial asymmetry in cleft lip and palate. *Cleft Palate Craniofac J* 1994;31:116–121
44. Ras F, Habets LL, van Ginkel FC, et al. Longitudinal study on three-dimensional changes of facial asymmetry in children between 4 to 12 years of age with unilateral cleft lip and palate. *Cleft Palate Craniofac J* 1995;32:463–468
45. Kuijpers MA, Desmedt DJ, Nada RM, et al. Regional facial asymmetries in unilateral orofacial clefts. *Eur J Orthod* 2015;37:636–642
46. Yang L, Chen Z, Zhang X. A cone-beam computed tomography evaluation of facial asymmetry in unilateral cleft lip and palate individuals. *J Oral Sci* 2016;58:109–115
47. Stauber I, Vairaktaris E, Holst A, et al. Three-dimensional analysis of facial symmetry in cleft lip and palate patients using optical surface data. *J Orofac Orthop* 2008;69:268–282
48. Starbuck JM, Ghoneima A, Kula K. A multivariate analysis of unilateral cleft lip and palate facial skeletal morphology. *J Craniofac Surg* 2015;26:1673–1678
49. Ahmad Y, Starbuck JM. Disruption of symmetry: a quantitative assessment of facial skeleton anatomy in children born with unilateral cleft lip and palate. *Clin Anat* 2018;31:1129–1136
50. Nkenke E, Lehner B, Kramer M, et al. Determination of facial symmetry in unilateral cleft lip and palate patients from three-dimensional data: technical report and assessment of measurement errors. *Cleft Palate Craniofac J* 2006;43:129–137
51. Abuhijleh E, Aydemir H, Toygar-Memikoğlu U. Three-dimensional craniofacial morphology in unilateral cleft lip and palate. *J Oral Sci* 2014;56:165–172
52. Zemann W, Santler G, Kärcher H. Analysis of midface asymmetry in patients with cleft lip, alveolus and palate at the age of 3 months using 3D-COSMOS measuring system. *J Craniomaxillofac Surg* 2002;30:148–152
53. Burstone CJ, James RB, Legan H, et al. Cephalometrics for orthognathic surgery. *J Oral Surg* 1978;36:269–277

Orbitofrontal Reconstruction With a Three-Dimensional Titanium Patient-Specific Implant After Intraosseous Haemangioma Resection

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Background: Intraosseous hemangiomas are rare benign tumors that can develop in the skull. Orbitofrontal localization is uncommon. The complexity of orbitofrontal anatomy results in difficult reconstruction following trauma or tumoral resection.
Case presentation: A 50-year-old woman with a right orbitofrontal intraosseous hemangioma was referred to our department. The authors decided to perform tumoral bone resection and orbitofrontal reconstruction using virtual surgical planning in collaboration with Materialize engineers (Materialise, Leuven, Belgium). Three cutting guides and a patient-specific 3-dimensional (3D) titanium implant were designed and manufactured in 3 weeks. Surgery was performed with a double surgical team composed of maxillofacial surgeons and neurosurgeons. No perioperative or post-operative complications occurred. Post-operatively, the patient was completely asymptomatic and clinical examination showed symmetrical and satisfactory facial morphology.

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