Correlation Between Soft and Hard Tissue Changes in the Zygomaticomaxillary Region After Bone Contouring Surgery for Fibrous Dysplasia—A Preliminary Study

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Purpose: The purpose of the present study was to determine the correlation between the soft and hard tissue changes in the zygomaticomaxillary region after facial bone contouring surgery for patients with craniofacial fibrous dysplasia (FD).

Materials and Methods: The present study was a retrospective case series that reviewed the cases of 13 patients with craniofacial FD in the zygomaticomaxillary region who had undergone navigation-guided facial bone contouring surgery from January 2013 to October 2017. Pre- and postoperative computed tomography (>3 months) were collected. The pre- and postoperative soft and hard tissues were placed in the same spatial coordinate system using multipoint registration to measure the distances between the corresponding pre- and postoperative points of the soft and hard tissues. The outcome variable was the corresponding soft tissue change. The correlation between the hard and soft tissue changes was obtained using correlation analysis with SPSS software (IBM Corp, Armonk, NY). The linear regression equation of the soft and hard tissue changes was used to predict the corresponding soft tissue changes.

Results: The Pearson correlation coefficient of the zygomatic region was 0.954 ($P < .001$) and the coefficient for the maxillary region was 0.758 ($P < .001$). The linear regression index (R^2) for the zygomatic and maxillary regions was 0.910 ($P < .001$) and 0.575 ($P < .001$), respectively. The β value of the linear regression equation for the zygomatic and maxillary regions was 0.815 ($P < .001$) and 0.52 ($P < .001$), respectively.

Conclusions: The soft and hard tissue changes were highly correlated in both the zygomatic area and the maxillary area, and the variance of the maxillary area was slightly greater than that in the zygomatic area. This implied that the change of 1 mm of bone tissue along the tangent direction of the bone contour will cause a change of 0.815 mm in the soft tissue in the zygomatic region and 0.52 mm in soft tissue in the maxillary region.

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Bone recontouring surgery seeks to reestablish a harmonious and symmetrical facial appearance and requires quantitative data regarding the association between soft and hard tissues to achieve accurate prediction of postoperative soft tissue changes.[1,2] The correlation between the soft and hard tissue changes of maxillary and mandibular repositioning after orthognathic surgery have been properly established from previous studies.[3-5] The delicate anatomic structures of the zygomaticomaxillary region will determine the width and prominence of the face and are responsible for the shape of the lateral segment of the middle third of the face.[6] However, to date, less attention has been given to the relationship between the soft and hard tissue changes in the zygomaticomaxillary region.

Fibrous dysplasia (FD) is a benign bone lesion characterized by the replacement of normal bone structure with abnormal fibro-osseous connective tissue. This was originally described by Lichtenstein in 1938 and Lichtenstein and Jaffe in 1942.[7] FD can be observed in 1 bone (monostotic) or multiple bones (polyostotic), and can be associated with endocrinopathies (McCune-Albright syndrome)[7] or intramuscular myxoma (Mazabraud's syndrome).[8] It represents ~2 to 3% of bone-derived tumors and 5 to 7% of benign bone tumors,[9] often occurring throughout the skeleton around the body, with a preference for the craniomaxillofacial area.[10,11] It will frequently involve the maxilla, mandible, frontal bones, sphenoidal bones, ethmoidal bones, parietal bones, temporal bones, and occipital bone, in descending order.[12] The maxilla alone, or with adjacent bones (eg, the zygoma), has been the most commonly affected site.

The natural history and clinical presentation of FD largely depends on the anatomic region involved. Regarding the craniomaxillofacial region, the most frequent clinical sign has been the painless, slowly enlarging, and hard swelling of the affected bone, resulting in facial asymmetry, disfigurement, malocclusion, orbital dystopia, and exophthalmos.[13] Malignant transformation of FD has been very rare, with the reported prevalence ranging from 0.4 to 4%.[14-16] Hence, the management principle for craniofacial FD should be restoration to a more esthetically satisfying and symmetrical facial appearance and correction of functional dysfunction.[17] For FD involving the zygomaticomaxillary region, facial bone contouring surgery will be the less invasive and relatively appropriate intervention, and the prognosis has been confirmed by a previous study,[18] which provided an excellent model for studying the soft and hard tissue changes in the zygomaticomaxillary region.

The purpose of the present study was to quantitatively determine the amount of soft tissue changes in response to bone contouring surgery for FD in the zygomatic and maxillary regions. The specific aim was to determine the ratio of hard to soft tissue changes after bony contouring surgery in FD to provide a reference for precisely predicting the soft tissue changes of similar cosmetic bone contouring surgery. We hypothesized that a strong correlation would be present between the soft and hard tissue changes in zygomaticomaxillary region.

Materials and Methods

PATIENT DEMOGRAPHIC DATA

The present study was a retrospective case series. We retrospectively reviewed the cases of 13 patients (6 males and 7 females; median age, 20 years) with craniofacial FD in the zygomaticomaxillary region who had undergone facial bone contouring surgery in Peking University School and Hospital of Stomatology (Beijing, China) from July 2013 to October 2017. The inclusion criteria were as follows: *1)* the surgical area involved the zygomaticomaxillary region; *2)* pre- and postoperative computed tomography (CT) scans were available with follow-up for 3 months or longer; and *3)* the patients had a pathologic diagnosis of FD. The exclusion criteria were as follows: *1)* the presence of craniofacial disorders, such as cleft lip and palate; *2)* a history of fracture in the facial area; and/or *3)* the presence of medical, physical, or mental conditions that could affect the patient's healing potential. All the patients met the inclusion criteria. The follow-up period ranged from 3 to 20 months (mean, 6.4; Table 1). The ethics committee of Peking University School and Hospital of Stomatology approved the present study (approval no. PKUSSIRB-201837100), and all the participants provided written informed consent.

The facial bone contouring surgery of all cases was performed according to the virtual plan and under the guidance of the computerized navigation system (BrainLAB AG, Feldkirchen, Germany). Before surgery, preoperative maxillofacial non–contrast-enhanced CT scans with a 1-mm slice thickness were acquired (field of view, 20 cm; pitch, 1.0; slice, 0.75 mm; 120 to 280 mA). The CT data in Digital Imaging and Communications in Medicine (DICOM) format were imported into iPlan (BrainLAB) software. The nonaffected side was mirrored to form the ideal contour and guide the surgical excision as a template. After surface registration, the intraoperative navigation was used to implement the virtual plan for bone contouring. During surgery, dysplastic zygomaticomaxillary bone was exposed using a combination of a transconjunctival preseptal approach, maxillary gingivobuccal approach, and supratarsal fold incision. A reduction

Table 1. CLINICAL DATA

Pt. No.	Age (yr)	Gender	Affected Side	Affected Area	Follow-Up Interval (mo)
1	18	Male	Right	Maxilla, zygoma, frontal bone, parietal bone, occipital bone	4
2	15	Male	Right	Maxilla, zygoma	4
3	17	Female	Right, left	Maxilla, zygoma, frontal bone, mandibular (left); zygoma (right)	3
4	29	Female	Left	Maxilla, zygoma, sphenoid bone, frontal bone, parietal bone, nasal bone, ethmoid	8
5	21	Male	Right	Maxilla, zygoma, sphenoid bone	4
6	23	Male	Left	Maxilla, zygoma	5
7	17	Male	Right	Maxilla, zygoma	3
8	27	Female	Left	Maxilla, zygoma, mandibular	7
9	17	Male	Right	Maxilla, zygoma	3
10	18	Female	Right	Maxilla, zygoma, sphenoid bone, ethmoid, mandibular	20
11	28	Female	Left	Maxilla, zygoma, temporal bone	5
12	44	Female	Right	Maxilla, zygoma	3
13	20	Female	Right, left	Maxilla, zygoma, temporal bone, parietal bone, mandibular (left), frontal bone	14

Abbreviation: Pt. No., patient number.

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of the burring of dysplastic bone was accomplished using a contouring burr to achieve symmetry.

MATERIALS AND METHODS

Step 1: Process of CT Data

A spiral CT scan was performed for each patient preoperatively and at least 3 months postoperatively (helix, with 1.25 mm slice thickness; BrightSpeed 16-slice CT scanner, GE Healthcare, Buckinghamshire, UK). The pre- and postoperative CT data in the DICOM format were imported to ProPlan CMF software (Materialise NV, Leuven, Belgium). Next, the hard tissue (HU threshold, 226 to 3071) and skin tissue (HU threshold, −718 to −177) were segmented and separately converted into STL file format. Subsequently, 4 STL files (preoperative hard tissue, preoperative soft tissue, postoperative hard tissue, postoperative soft tissue) were imported into the ProPlan CMF software.

Step 2: Registration and Superimposition

To diminish the bias created by the different patient position strategies during the CT scans, the head position of each scan was manually oriented, such that the Frankfurt horizontal (FH) plane was connected to the orbitale of the unaffected side, and the porion of both sides was parallel to the ground. Because the cranial base structures were stable and could not be altered by surgery, 9 hard tissue landmarks (nasion, orbitale right, orbital left, porion, anterior nasal spine [ANS], menton, gonion right, gonion left, basion) of the cranial base and the unaffected side were used as the superimposition reference for the pre- and postoperative hard tissue STL files. The entire process was achieved using automatic multipoint registration and manual adjustments. The pre- and postoperative soft tissue STL files were then manually paired to the hard tissue STL files. Simultaneously, the contour line of each STL file was shown on 1 axial CT scan of the same plane parallel to the FH plane (Fig 1).

The accuracy of the superimposition was then evaluated using color-coded map analysis, which revealed the distance between the pre- and postoperative registered surface models (Fig 1). The green color-coded areas represented surface distances of less than 6 mm, which could be seen on the cranial base of the unaffected side, indicating adequate superimposition. Some areas with larger surface distances (shown in gray) indicated the surgical area.

Step 3: Measurement

The surgical area was divided into the zygomatic and maxillary regions by the plane through the ANS point parallel to the FH plane. After superimposition of the STL files of the preoperative soft tissue, postoperative soft tissue, preoperative bone tissue, and postoperative bone tissue, the surgical site had 4 different contour lines. The first axial CT slice showing an intact zygomatic arch on both sides was chosen as the first slice for the measurements. From the anterior border to the posterior border of the surgical site, 3 points dividing the preoperative bone contour into 4 contour lines with equal length were made on the same axial CT slice. Using the same method, 6 more points were selected on the 2 neighboring slices of the axial

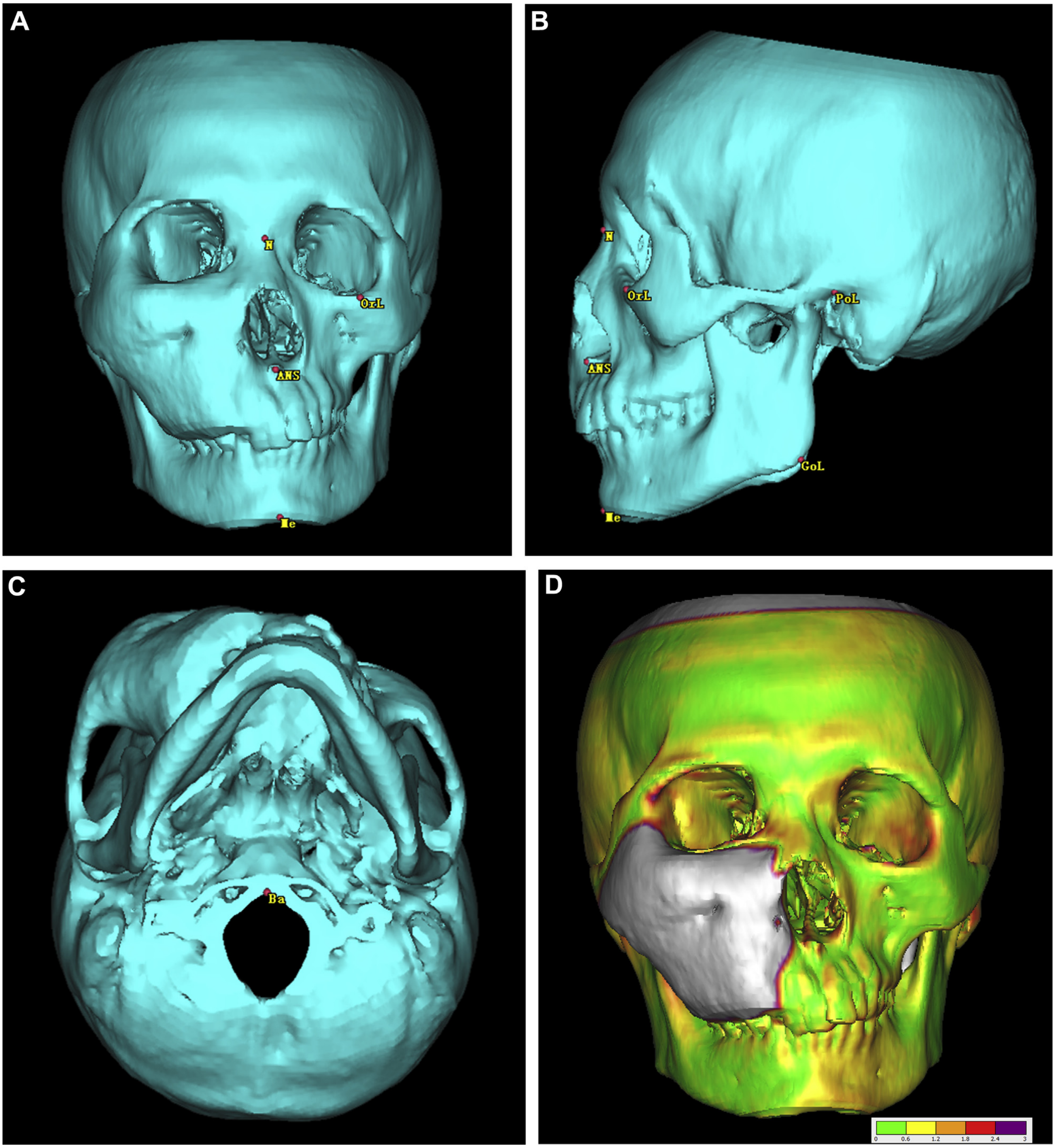

FIGURE 1. Registration and superimposition of pre- and postoperative soft and hard tissue STL files. *A-C*, Nine hard tissue landmarks for multi-point registration: *A*, anterior view; *B*, lateral view; *C*, inferior view. *D,E*, Color-coded map to evaluate the accuracy of registration: *D*, anterior view; **(Fig 1 continued on next page.)**

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CT plane by the same clinician to diminish the bias. Nine points were made to reflect the hard and soft tissue changes of each area. Through each point, a tangent line of the arc of the preoperative bone contour was made. A reference plane was then made perpendicular to the tangent line through each point, which intersected the 4 contour lines (preoperative bone, postoperative bone, preoperative skin, and postoperative skin) at the points of bone 1, bone 1′, skin 1, and skin 1′ (Fig 2). Along the reference plane, the shortest distances between the points of bone 1 and bone 1′ and the points of skin 1 and skin 1′ were

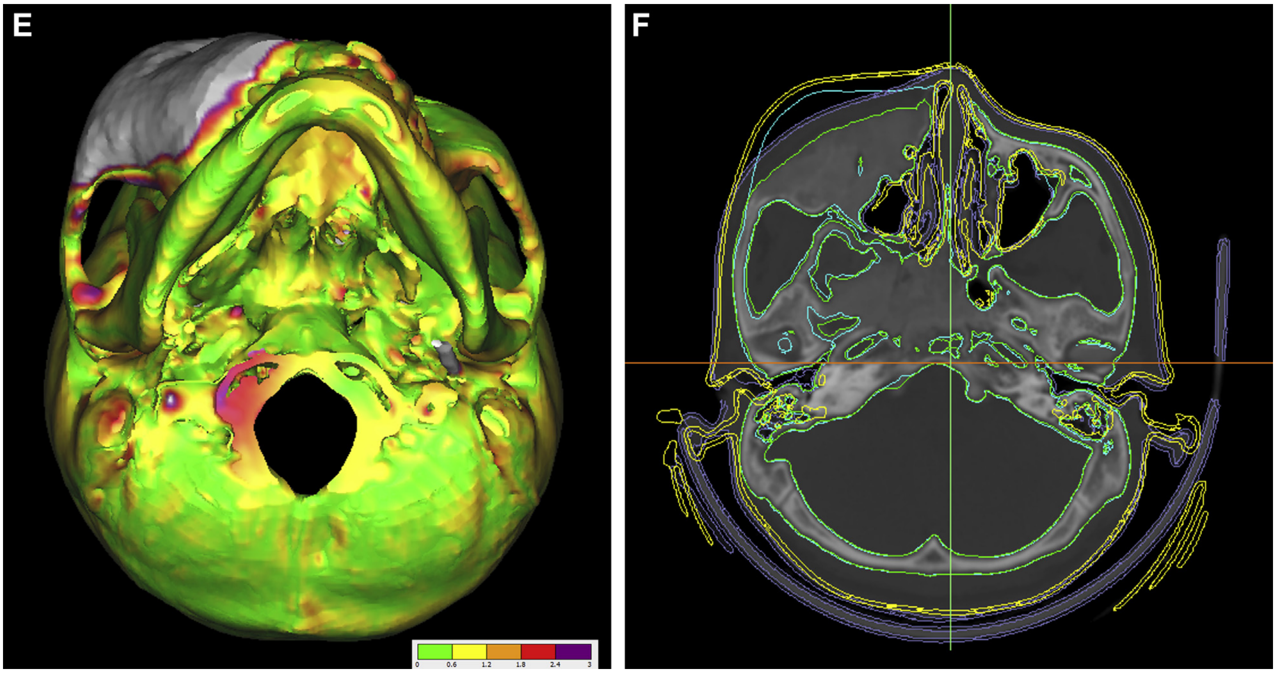

FIGURE 1 (cont'd). *E*, inferior view. *F*, After superimposition of soft and hard tissue STLs, the contour line of the STLs was shown in 1 axial computed tomography scan. The *blue*, *green*, *yellow*, and *purple* represent the preoperative hard tissue, postoperative hard tissue, preoperative soft tissue, and postoperative hard tissue, respectively.

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measured using the cephalometric analysis module of ProPlan CMF. The average value of the 9 points of each patient for each anatomic region was used for statistical analysis.

Step 4: Statistical Analysis

Data management and analysis were performed using SPSS, version 24.0, software (IBM Corp, Armonk, NY). The patients were divided into 2 age groups by the median age. The difference in the 2 genders and age groups were tested using the 2-sample t test. The Pearson correlation coefficients for the hard and soft tissue changes in the zygomatic and maxillary regions were calculated and tested against the null hypothesis that the Pearson correlation coefficient would be 0. A general linear model was used to predict the soft tissues changes, with only hard tissue changes alone included as a predictor. A 2-sided value of $P < .05$ was considered statistically significant.

Results

All 13 patients had met the inclusion criteria. The follow-up period ranged from 3 to 20 months (mean, 6.4 months; Table 1; Fig 3). The average hard and soft tissue changes in both regions of each patient are presented in Table 2. No statistically significant differences were found in the soft tissue changes between the 2 genders or the age groups in either region (Table 3). The Pearson coefficient of hard and soft tissue changes in the zygomatic region was 0.954 ($P < .001$) and in maxillary region was 0.758 ($P < .001$). The results of linear regression are shown in Table 4. The linear regression index (R^2) of the linear model using only the hard tissue change in the zygomatic and maxillary regions was 0.910 ($P < .001$) and 0.575 ($P < .001$), respectively. The linear regression equations are shown in Figures 4 and 5. The β value obtained from the linear regression equation in the zygomatic and maxillary regions was 0.815 and 0.520, respectively. Of the surgical area above the ANS point, a 1-mm change in the bone tissue along the tangent direction of the bone contour will cause a change of 0.815 mm in the soft tissue, with a statistically significant difference. Of the surgical area below the ANS point, a 1-mm change of bone tissue along the tangent direction of the bone contour will cause a change of 0.52 mm in the soft tissue, with a statistically significant difference.

Discussion

The arrangement of facial soft tissues is important for oral and maxillofacial surgeons. The malar prominence contributes largely to the overall facial appearance, and the zygomatic region is considered one of the most esthetic areas for establishing facial contour.[19] Volume or contour deficiencies of the midface

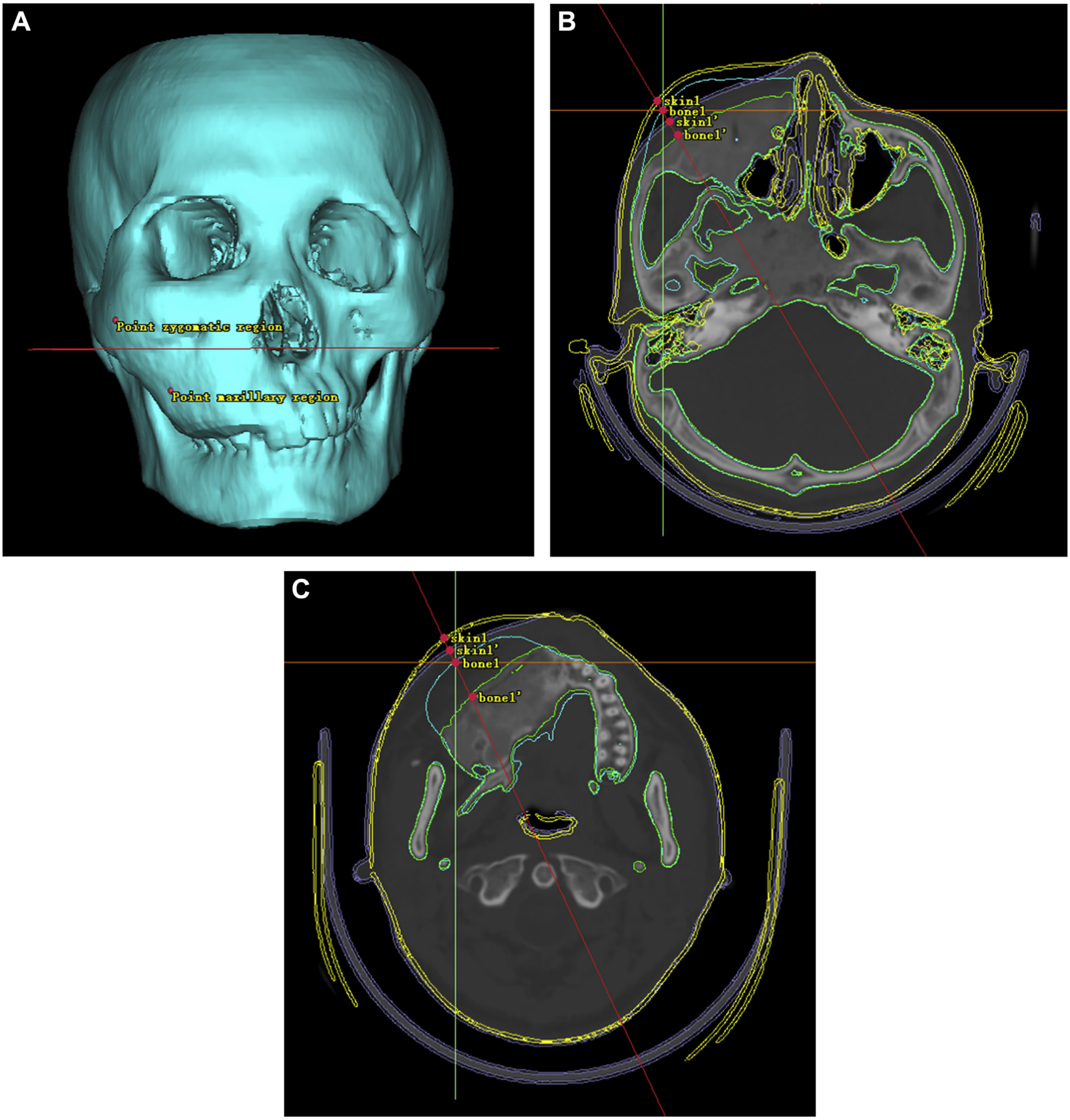

FIGURE 2. Measurements of soft and hard tissue changes between pre- and postoperative computed tomography scans. *A,* The surgical area was divided into the zygomatic and maxillary region by the plane through the anterior nasal spine point parallel to Frankfort horizontal plane. Each representative point of each region was determined. *B,C,* A reference plane (*red*) was made perpendicular to the tangent line through each quarter-point of the surgical area (ie, point bone 1), which intersected the 4 contour lines at point bone 1, bone 1′, skin 1, skin 1′: *B,* zygomatic region; *C,* maxillary region.

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due to congenital disease,[20] trauma,[21] and simply aging[22,23] have recently been appreciated as causes of facial esthetic imbalance. The management approach for malar deficiency has ranged from silicone implants[21,22,24] to osteochondral grafting[25,26] and shares the same principle for recontouring the hard tissue to achieve facial symmetry, implying that the soft/hard tissue relationship of the malar region will directly determine the surgical effect. However, the available studies failed to establish an oblique soft/hard tissue relationship to determine the midfacial prominence, which is the dominant concern of

FIGURE 3. *A,B,* Preoperative views and *C,D,* 3-month postoperative views of a patient with fibrous dysplasia in the right zygomaticomaxillary region.

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malar augmentation procedures and bone contouring surgery in the midfacial area. Hence, the purpose of the present study was to quantitatively determine the amount of soft tissue changes in response to bone contouring surgery for FD at the zygomatic and maxillary regions. Our specific aim was to determine the ratio of hard to soft tissue changes after bony contouring surgery in FD to provide a reference to precisely predict the soft tissue changes of similar cosmetic bone contouring surgery.

Because of the relatively constant surgical area and similar cosmetic management principles with the malar augmentation procedure, the facial bone contouring surgery for craniofacial FD in the

Table 2. AVERAGE PRE- AND POSTOPERATIVE HARD AND SOFT TISSUE CHANGES OF ZYGOMATICOMAXILLARY REGION

	Zygomatic Region		Maxillary Region	
Pt. No.	ΔHard Tissue (mm)	ΔSoft Tissue (mm)	ΔHard Tissue (mm)	ΔSoft Tissue (mm)
1	18.5	14.7	16.1	9.3
2	6.0	4.1	12.0	4.8
3	6.8	3.2	8.0	2.8
4	12.3	10.2	8.3	7.8
5	20.4	15.4	21.8	13.4
6	6.3	3.8	9.2	5.1
7	9.0	5.0	11.1	4.6
8	11.5	5.2	7.7	6.5
9	16.2	11.2	14.4	5.5
10	118	9.9	11.1	8.6
11	7.4	4.7	10.1	4.5
12	4.6	4.2	NA	NA
13	9.1	5.5	7.5	4.8

Abbreviations: NA, not applicable (surgical area did not involve maxillary region); Pt. No., patient number.

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zygomaticomaxillary region served as an excellent model in the present study to quantitatively analyze the soft tissue changes that occur after the hard tissue changes resulting from facial bone contouring surgery. We hypothesized that a strong correlation would be present between the soft and hard tissue changes in the zygomaticomaxillary region.

The present retrospective study included 13 patients with a diagnosis of FD involved in zygomaticomaxillary area who had undergone facial bone contouring surgery with at least a 3-month follow-up period. According to the computed tomography scans, none of the patients had experienced a recurrence of FD during the follow-up period. In the present study, a significant variation between the correlation of hard and soft tissue changes in the zygomatic region (R^2 = 0.910) with the correlation in the maxillary region (R^2 = 0.575) showed varying characteristics of the soft/hard tissue relationship in the zygomaticomaxillary region. This confirmed our hypothesis that a strong correlation exists between the soft and hard tissue changes in the zygomaticomaxillary region. Although the slope of linear regression of the maxillary region is not as steep as that in the zygomatic region, the soft tissue changes were still predictable. This finding supports evidence reported by previous studies that the anterior segment (including the infraorbital rim and the most anterior projection of the malar bone) and the lateral segment (including the remainder of the malar eminence, lateral orbital

Table 3. COMPARISON OF SOFT TISSUE CHANGES IN 2 REGIONS STRATIFIED BY GENDER AND AGE

Region	Covariate	Mean ± Standard Error (mm)	*t* Value	*P* Value
Zygomatic	Gender		1.257	.235
	Male	9.0 ± 5.4		
	Female	6.1 ± 2.9		
	Age group		0.156	.878
	≤20 yr	7.6 ± 4.3		
	>20 yr	7.3 ± 4.6		
Maxillary	Gender		0.769	.459
	Male	7.1 ± 3.5		
	Female	5.8 ± 2.2		
	Age group		−1.007	.338
	≤20 yr	5.8 ± 2.3		
	>20 yr	7.5 ± 3.6		

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Table 4. LINEAR REGRESSION ANALYSIS OF HARD AND SOFT TISSUE CHANGES IN THE ZYGOMATICO-MAXILLARY REGION

	Hard Tissue Change			
Region	β	95% CI	*P* Value	R^2
Zygomatic	0.815	0.644-0.985	<.001	0.910
Maxillary	0.520	0.205-0.834	<.001	0.575

Abbreviation: CI, confidence interval.

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rim, and zygomatic arch) should be separately evaluated because recontouring along these 2 different directions can lead to a distinctly different appearance.

The observed variation in the soft/hard tissue correlation between the zygomatic region and the maxillary region might be explained by the different soft tissue thicknesses determined by the anatomic structures. A large-scale study of the facial soft tissue depths of white adults showed that the soft tissue of the zygomatic eminent zone is much thinner than the inferior malar zone.[27] In accordance with the present results, previous studies have demonstrated that the preoperative soft tissue thickness can lead to significantly different soft/hard tissue ratios after Le Fort I surgery.[28-30] Several studies of the contribution of subcutaneous fat to the face found that the malar fat is composed of 3 separate compartments: the medial, middle, and lateral temporal cheek fat.[31,32] A study of white adults also showed that the soft tissue depth of the zygomatic eminent zone was close to the soft tissue depth at the menton point, implying that the soft/hard tissue ratio of the zygomatic region (0.815:1) was close to the soft/hard tissue ratio of the chin procedure (0.9:1).[33] The mechanism associated with the clinical finding that thinner soft tissue has a stronger correlation with hard tissue changes and higher soft/hard tissue ratios is still ambiguous. A possible explanation for this might be that the compartments are composed of a pocket of air between those fat pads that absorb the trimming of the maxilla, which can serve as a “buffer” to the hard tissue changes. Further study is required to explain whether the soft tissue thickness has a great influence on the soft/hard tissue relationship.

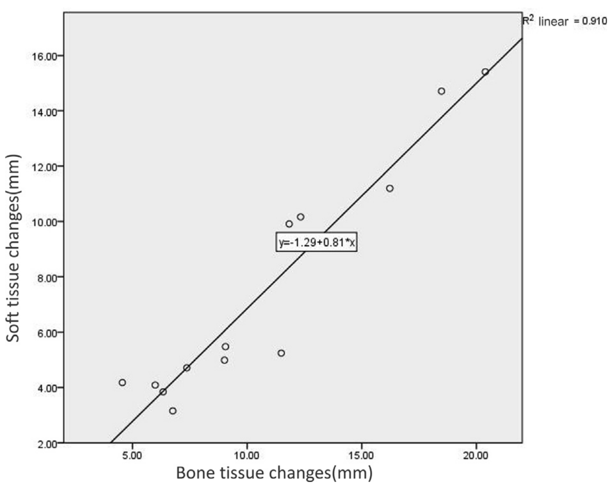

FIGURE 4. Linear regression graph of hard and soft tissue changes in the zygomatic region.

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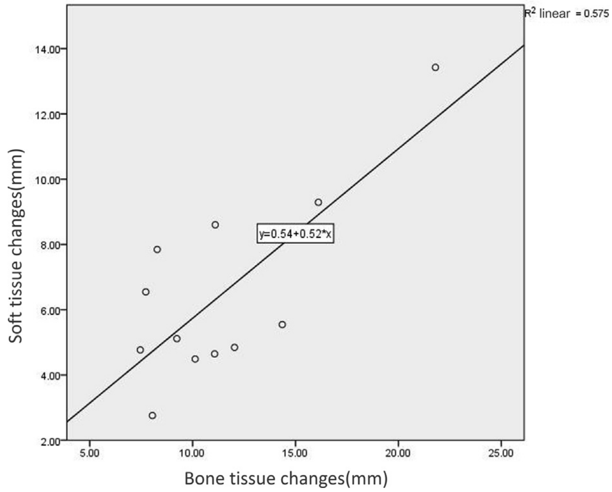

FIGURE 5. Linear regression graph of hard and soft tissue changes in the maxillary region.

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To diminish the influence of postoperative swelling, the present study only included those patients with at least a 3-month follow-up period. The data derived from several studies,[34-36] which had mainly concentrated on the postoperative swelling after orthognathic surgery, suggested that only 20% of the initial edema remained and facial morphology had recovered to ~83 to 90% of the baseline facial scan after 3 months. Soft and hard tissue analyses used the CT scans, whose accuracy of measuring soft tissue thickness of facial region has been confirmed by a previous study.[37] For the purposes of measuring the soft and hard tissue changes after surgery, we used the cranial base as a superimposition reference, for which the associated accuracy and reproducibility were confirmed by Nada et al.[38] Several investigators have used 3-dimensional (3D) measurements to analysis the soft/hard tissue ratio in orthognathic surgery, and superimposition is the most well-known tool for the registration and measurement of pre- and postoperative data.

Other factors that can contribute to the variability of the values of the soft/hard tissue ratios included the interval time, weight and facial expressions of the patient, accuracy of the superimposition of the 3D models, and the precise definition and location of landmarks. Soft tissue changes with remodeling of the underlying skeleton can most certainly continue for longer than 3 months postoperatively. Hence, the results of the present study are preliminary and should be cautiously assessed.

In conclusion, the present retrospective study revealed a correlation between the soft and hard tissue changes after conservative surgery for patients with craniofacial FD in the zygomaticomaxillary area. In the zygomatic region, the soft and hard tissue changes show a significant linear relationship, with a soft/hard tissue ratio of 0.815:1, implying that removal of 1 mm of bone tissue along the tangent direction of the bone contour will cause a change in the soft tissue of 0.815 mm soft tissue, with a statistically significant difference. In the maxillary region, the soft and hard tissue changes showed a poor correlation with a soft/hard tissue ratio of 0.52:1, demonstrating that the removal of 1 mm of bone tissue along the tangent direction of the bone contour will cause a change of 0.52 mm in the soft tissue. The present study has provided the first quantitative analysis of the soft and hard tissue changes in the zygomaticomaxillary area, laying a foundation for future research regarding the preoperative prediction of soft tissue changes and the precise reconstruction of the zygomaticomaxillary area combined with a digital surgical navigation system. Future work focusing on the long-term soft tissue changes with a longer follow-up interval is required.

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