A Quantitative Three-Dimensional Analysis of Coronoid Hypertrophy in Pediatric Craniofacial Malformations

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Background: Coronoid process hypertrophy can be associated with a variety of congenital or acquired anomalies. There is, however, no consensus on a quantitative or objective measure to define coronoid hypertrophy. Here, the authors describe a novel analytical technique using three-dimensional computed tomographic data to accurately and reproducibly assess coronoid size and diagnose coronoid:condyle disproportion.
Methods: A total of 24 patients were analyzed using three-dimensional medial axis analysis, eight with of unilateral coronoid hypertrophy, four with of bilateral coronoid hypertrophy, and 12 age-matched normal control patients.
Results: Measurement of normal subjects ($n = 12$) demonstrated a coronoid:condyle *volumetric* ratio less than or equal to 0.5. Analysis of patients with coronoid hypertrophy demonstrated that a coronoid:condyle volumetric ratio greater than or equal to 1.0 was consistent with marked coronoid:condylar disproportion and a ratio between 0.5 and 1.0 was indicative of modest disproportion. *Surface area* ratios comparing coronoid with condyle were also elevated (ratio, ≥0.5) in patients with coronoid hypertrophy.
Conclusions: Quantitative assessment of coronoid size using three-dimensional volume and surface area analysis of computed tomographic data may be helpful to the clinician in diagnosing coronoid hypertrophy and in guiding treatment. It may also serve a role in monitoring the temporal evolution of coronoid hypertrophy in early cases that have not yet resulted in trismus or decreased interincisal opening. (*Plast. Reconstr. Surg.* 129: 312e, 2012.)
CLINICAL QUESTION/LEVEL OF EVIDENCE: Diagnostic, IV.

During normal mouth opening, the temporalis muscle relaxes so that the condyle can rotate and translate in the glenoid fossa. However, temporalis muscle strain across an akylosed temporomandibular joint or hyperactive temporalis activity across a normal joint can stimulate osteogenesis, causing coronoid process elongation. Ultimately, the resulting coronoid hypertrophy can restrict condylar movement within the temporomandibular joint and limit mandibular excursion (Fig. 1).

Coronoid hypertrophy is a rare condition in children.[1,2] The etiology of coronoid hypertrophy is not well-described, but it has been found in association with many conditions, including temporomandibular joint ankylosis, unilateral craniofacial microsomia, Treacher Collins syndrome, trismus, and temporalis overactivity.[3–5] Coronoid hypertrophy is often a difficult and subjective clinical diagnosis due to the variable degree of bony overgrowth.[5] Furthermore, because coronoid overgrowth can be associated with a variety of congenital or acquired anomalies, the relationship between the coronoid and the rest of the craniofacial skeleton can vary considerably.[1] Mandibular morphology has traditionally been analyzed by cephalometry, but this technique is unsuitable for young patients (<4 years old) due to limitations in head positioning, mandibular orientation, and patient compliance. To assess coronoid size, volume, and surface area in young patients, we developed a quantitative analytical technique using three-dimensional computed tomographic data.

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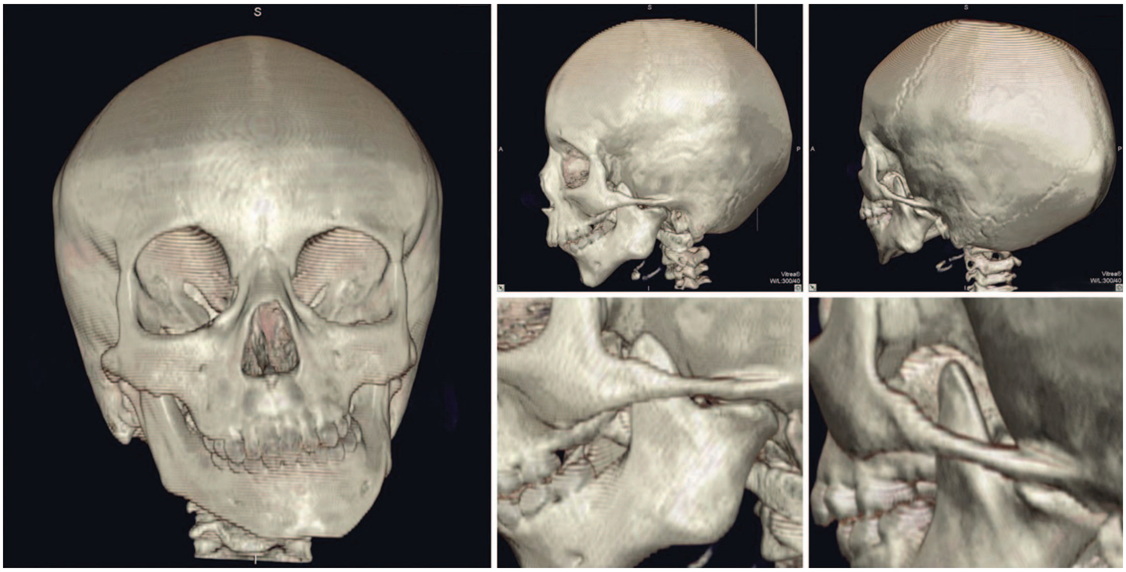

Fig. 1. Three-dimensional computed tomographic scan of a 4-year-old girl with left craniofacial microsomia and unilateral coronoid hypertrophy. Note that the large coronoid process extends above the zygomatic arch and abuts the posterior zygoma, impeding rotation and translation of the mandible. (*Left*) Frontal view; (*above, center and right*) lateral views; (*below, center and right*) oblique views.

PATIENTS AND METHODS

Cases and Controls

A retrospective review of 12 pediatric patients (mean age, 6.8 years; range, 23 months to 15 years) treated for unilateral or bilateral coronoid hypertrophy at the New York University School of Medicine, Institute of Reconstructive Plastic Surgery (Institutional Review Board no. 08-676), and at the Department of Maxillofacial Surgery of the Hospital de Salvador, Children's Hospital Exequiel González Cortés, Universidad Mayor, in Santiago, Chile, was performed. Case subjects were compared with 12 age-matched controls who underwent routine radiographic evaluation for trauma.

Three-Dimensional Computed Tomography and Medial Axis Analysis

All patients were scanned using a craniofacial computed tomographic protocol with three-dimensional reconstruction. Thin-section (1.5 to 3 mm) series were uploaded in Digital Imaging and Communications in Medicine file format to a dedicated image-reconstruction workstation (Vitrea version 2.0, Vital Images, Minnetonka, Minn.), and bone-window contrast settings were used during image reformatting and segmentation.

Analysis was performed bilaterally for normal controls and unilaterally in the case of coronoid pathology; in cases of bilateral coronoid hypertrophy, the left side was arbitrarily chosen for analysis. Three-dimensional medial axis analysis was performed to isolate the coronoid and condylar processes, based upon the two-dimensional cephalometric methodology previously described by the senior authors (J.G.M. and B.H.G.).[6] Briefly, reconstructed sagittal sections were oriented using the Frankfort horizontal plane, and a midsagittal plane through the mandibular ramus was established. This axial plane provided an *en face* view of the upper ramus, coronoid, and condyle that consistently gave the largest "silhouette" for analysis.

A series of intraosseous circles were inscribed within the borders of the coronoid, condyle, and mandibular ramus such that each circle was tangent to the osseous surface (Fig. 2, *above*). Lines connecting the centers of these circles defined a Y-shaped path, and the point of convergence of the arms defined the center of the principal circle used for further analysis. This principal circle was tangent to both the anterior and posterior borders of the mandibular ramus and to the nadir of the sigmoid notch (Fig. 2, *below*).

Chords connecting these points of tangency were drawn to objectively define the lower border of the coronoid process and mandibular condyle (Fig. 2, *below, right*), and these segments were projected medially and laterally to establish the planar "bases" of the coronoid and condylar "pyramids." Subsequently, the three-dimensional contour of the coronoid and condyle were segmented, with data outside these volumes of interest discarded to effectively isolate the coronoid and condyle (Fig. 3). Vol-

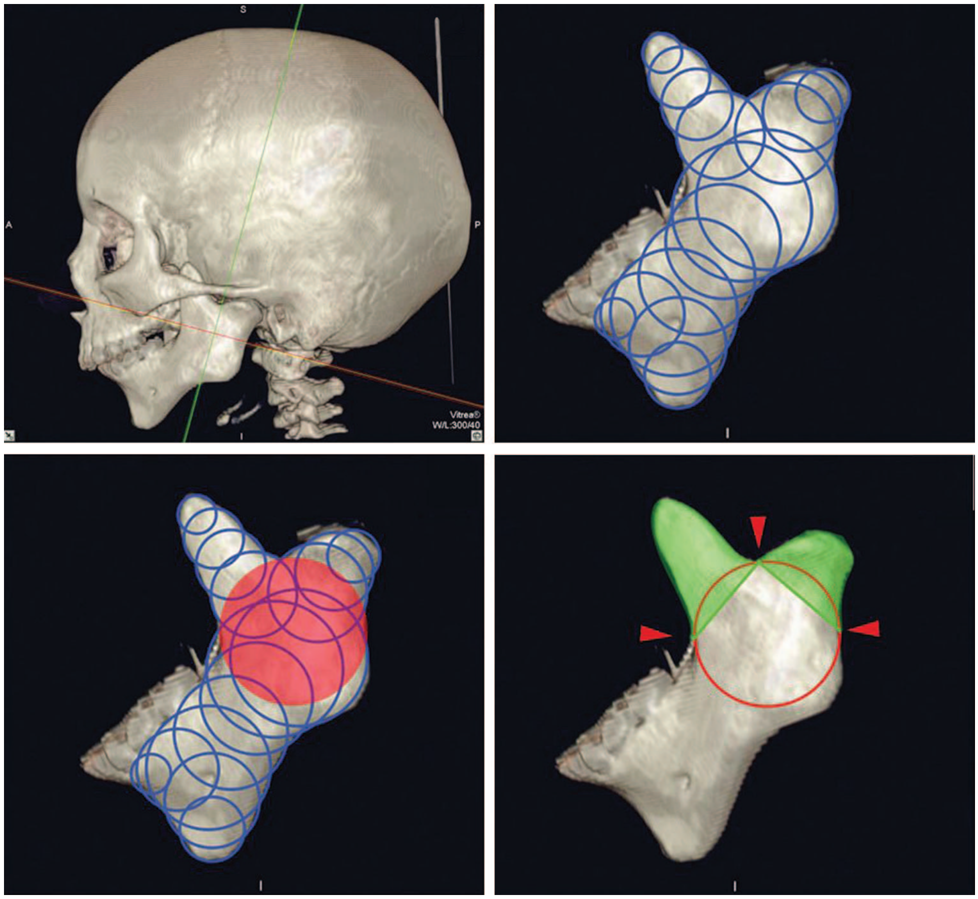

Fig. 2. Medial axis analysis was performed to isolate the coronoid and condylar processes. By using three-dimensional postprocessing software, the image was oriented to the Frankfurt horizontal, and a midsagittal plane bisecting the mandibular ramus was established (*above, left*). This provided an *en face* "silhouette" of the mandible. A series of intraosseous circles were inscribed within the coronoid, condyle, and mandibular ramus such that each was tangent to the bony edges (*above, right*). A principal intraosseous circle (*shaded*) was defined with tangents at the anterior aspect of the ramus, posterior aspect of the ramus, and nadir of the sigmoid notch (*below, left*). The base of the coronoid process was defined inferiorly by the chord connecting the anterior tangent point (*red arrows*) to the sigmoid notch tangent point, and the base of the condylar process was likewise defined by the chord connecting the posterior tangent point (*red arrows*) to the sigmoid notch tangent point (*below, right*).

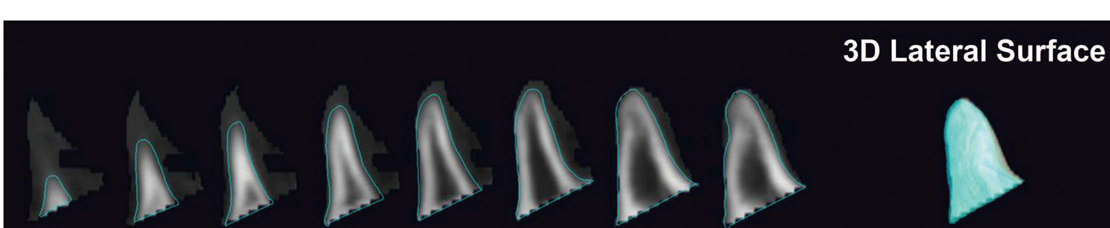

Fig. 3. After the coronoid and condylar processes were isolated, thin sections (1 mm) were used to measure the cross-sectional surface area and perimeter of each bony segment. A series of sections from a single coronoid process is shown with the region of interest outlined (*left*). The volume and surface area were calculated by summation of serial sections in the three-dimensional reconstructed view (*right*).

ume and surface area of each structure were calculated, and the coronoid:condyle volume and surface area ratios were calculated.

Statistical Analysis

Descriptive statistics and measurements were analyzed using statistical analysis software (SPSS version 14, SPSS Inc., Chicago, Ill.). Means were compared using a two-tailed *t* test for equality of means assuming unequal variances. Data are represented as mean ± SEM.

Table 1. Demographics

Class	Mean Age (yr)	*n*	Male	Female	SD
Case	6.77	12	5	7	3.75
Bilateral	6.48	4	2	2	3.15
Unilateral	6.92	8	3	5	4.21
Control	6.92	12	6	6	4.06

t Tests against ages: case versus control, $p = 0.93$; bilateral versus control, $p = 0.85$; unilateral versus control, $p = 1.00$; bilateral versusunilateral, $p = 0.86$.

RESULTS

Eight cases of unilateral and four cases of bilateral coronoid hypertrophy were identified. The diagnosis of coronoid hypertrophy was confirmed by expert review of computer tomographic studies by a pediatric neuroradiologist. The age range of the patients was 23 months to 15 years. Five cases were male and seven female. Twelve age-matched controls were identified for a total of 24 control hemimandibles (range, 1 to 14 years). Statistical comparison confirmed that the coronoid hypertrophy population and normal controls were appropriately age-matched ($p = 0.929$) (Table 1).

Coronoid Hypertrophy Is Variable

Analysis of the three-dimensional reconstructions revealed a wide array of phenotypic morphology (Fig. 4), ranging from a coronoid that is broad and wide to one that is tall and narrow. In addition, the coronoid–zygoma relationship was

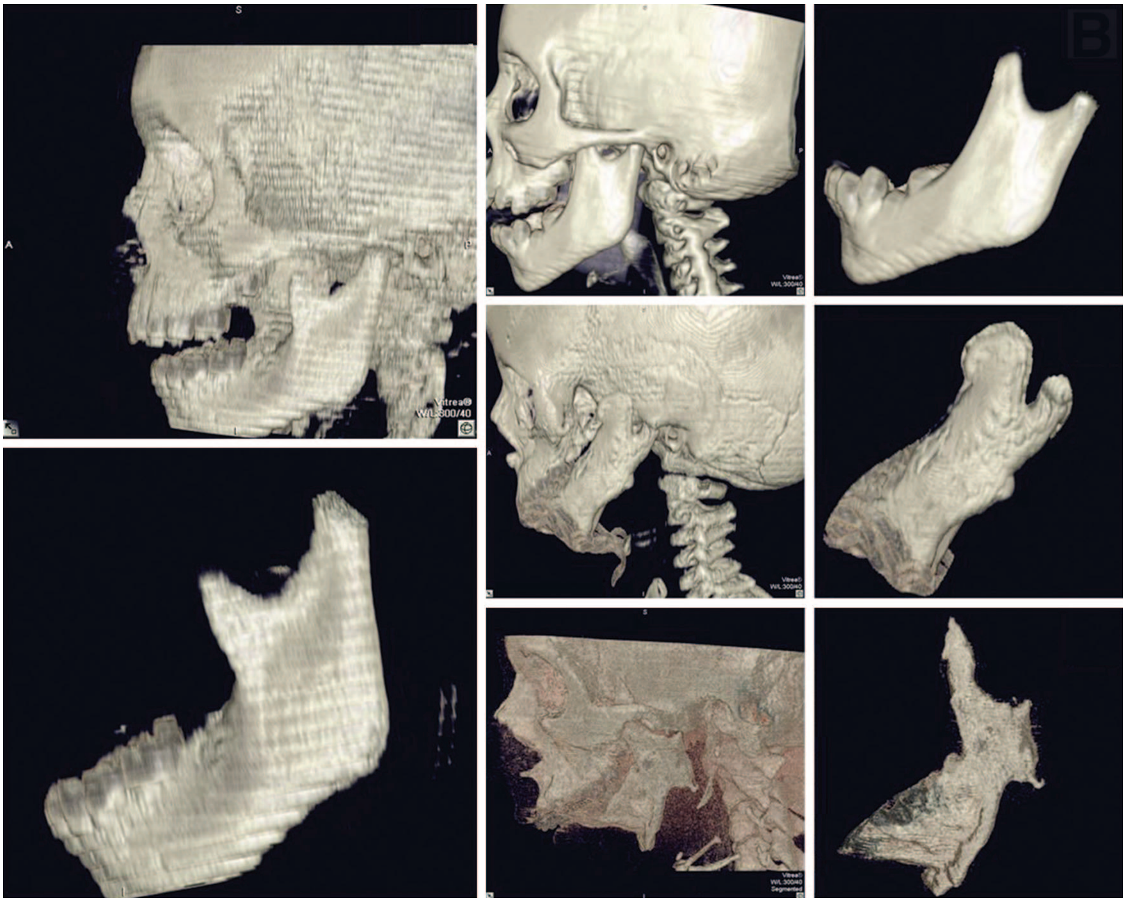

Fig. 4. The variable radiographic presentations of coronoid hypertrophy. Normal mandibular morphology consists of a smaller coronoid than the ipsilateral condyle (*left, above and below*, normal control case 1). Three pathologic mandibles with coronoid hypertrophy are also shown (*above, center and right*, case 11; *center, center and right*, case 1; *below, center and right*, case 5).

not uniform, because many patients had zygomatic pathology in addition to mandibular dysmorphology. A number of patients were not in centric occlusion at the time of their scans. Nevertheless, the mandibular coronoids and condyles were successfully segmented by the same methodology in all cases, irrespective of age, pathologic diagnosis, or orientation of the mandible.[6]

Coronoid:Condyle Disproportion Can Be Defined by an Increased Coronoid:Condyle Ratio

Raw measures of absolute coronoid and condylar volume and surface area were obtained; however, these values were not ideal for analysis because overall mandible size varied greatly by age and pathology. Table 2 highlights the high variance for absolute measurements of coronoid volume, coronoid surface area, condylar volume, and condylar surface area. In contrast, determination of the relative coronoid:condyle volume ratio and coronoid:condyle surface area ratio revealed tight clustering (especially for controls) that was suitable for comparison (Table 3; Fig. 5).

Normal controls had a mean volume ratio of 0.39 ± 0.02, compared with 1.29 ± 0.20 for coronoid hypertrophy patients. Similarly, the surface area ratio for normal controls was 0.42 ± 0.01, whereas the cases had a ratio of 1.08 ± 0.12. Significance tests confirm that these differences are significant ($p < 0.01$) for both volume and surface ratios.

Table 2. Raw Volume and Surface Area Measurements

	n	Minimum	Maximum	Mean	SD
Controls					
Coronoid volume	24	0.31	1.03	0.64	0.23
Coronoid SA	24	3.60	7.70	5.55	1.31
Condyle volume	24	0.83	2.70	1.67	0.61
Condyle SA	24	8.60	18.10	13.15	2.96
Volume ratio	24	0.26	0.51	0.39	0.075
SA ratio	24	0.30	0.58	0.42	0.058
Cases—bilateral					
Coronoid volume	16	0.50	10.70	3.06	3.37
Coronoid SA	16	0.97	16.80	7.29	4.33
Condyle volume	16	0.33	9.10	2.71	3.23
Condyle SA	16	1.50	12.80	6.44	3.48
Volume ratio	16	0.67	3.08	1.34	0.74
SA ratio	16	0.57	2.04	1.13	0.40
Cases—unilateral					
Coronoid volume	7	0.99	9.90	4.1986	3.39
Coronoid SA	7	1.20	16.80	7.6286	5.18
Condyle volume	7	0.79	9.10	3.7271	3.72
Condyle SA	7	1.50	9.70	6.1000	3.37
Volume ratio	7	0.75	3.08	1.5172	1.00
SA ratio	7	0.71	1.77	1.1938	0.34

SD, standard deviation; SA, surface area.

Table 3. Coronoid:Condyle Volume and Surface Area Ratios

	R Volume Ratio	R Surface Area Ratio	L Volume Ratio	L Surface Area Ratio
Cases				
1	1.96	2.04	1.25	1.20
2	1.20	1.04	—	—
3	0.67	0.63	—	—
4	1.16	1.24	—	—
5	—	—	3.08	1.37
6	1.18	0.57	1.10	1.27
7	1.39	1.01	2.83	1.77
8	0.76	0.97	—	—
9	—	—	1.10	1.27
10	1.82	1.37	2.58	1.85
11	0.67	0.81	0.75	0.87
12	—	—	0.75	1.17
Mean				
Age-matched controls				
1	0.26	0.35	0.30	0.38
2	0.29	0.40	0.32	0.42
3	0.39	0.41	0.45	0.36
4	0.37	0.44	0.33	0.43
5	0.46	0.42	0.49	0.43
6	0.43	0.45	0.43	0.47
7	0.49	0.56	0.51	0.58
8	0.37	0.41	0.41	0.45
9	0.40	0.41	0.39	0.42
10	0.48	0.43	0.47	0.42
11	0.32	0.41	0.32	0.41
12	0.27	0.30	0.41	0.43
Mean				

R, right; L, left.

Coronoid:Condyle Volume and Surface Area Ratios Greater than 0.5 Identify Disproportion

As expected, the data demonstrate that cases of coronoid hypertrophy had higher volume and surface area ratios. To establish a threshold of significance for a test of coronoid:condyle disproportion, we defined volume and surface area ratios that were greater than the mean ratio for controls plus two standard deviations. For the population studied, this calculation yielded a volume ratio more than 0.55 and a surface area ratio greater than 0.54. These values were both approximated as 0.50 to improve the simplicity and utility of the method.

Next, we determined the sensitivity and specificity of these tests using the study populations (Tables 4 and 5). Both tests had a sensitivity of 100 percent and high specificities (96 percent for volume ratio and 92 percent for surface area ratio). The positive predictive values, of volume and surface area ratios greater than 0.5, were 92 percent

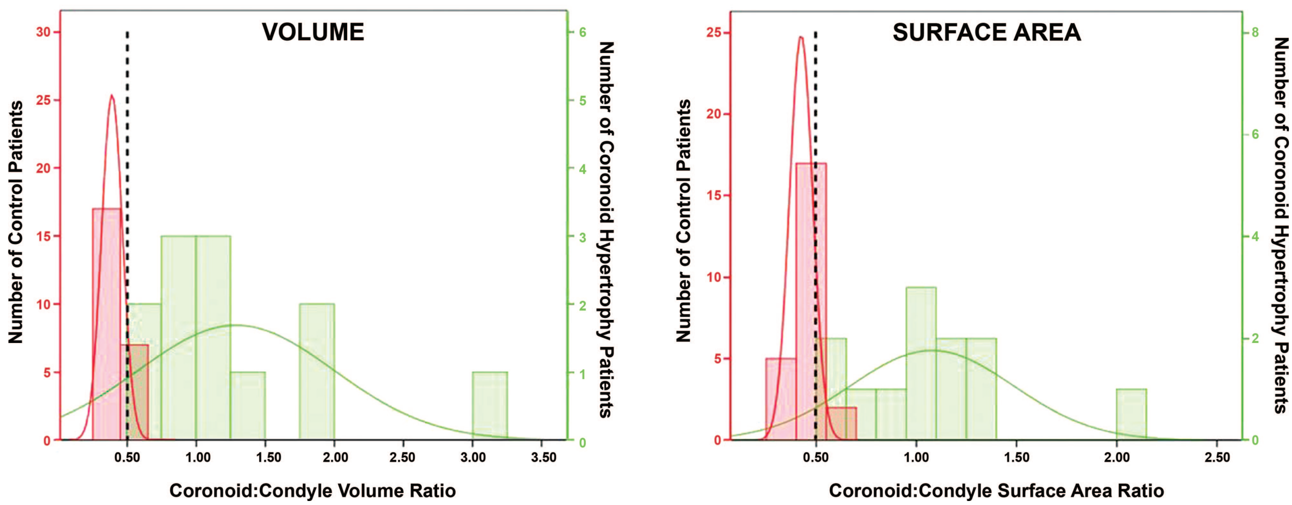

Fig. 5. (*Left*) Patients with coronoid hypertrophy (*red*) have significantly elevated coronoid:condyle volume ratios ($p < 0.01$) when compared with age-matched controls (*green*). (*Right*) Similarly, patients with coronoid hypertrophy have significantly elevated coronoid:condyle surface area ratios ($p < 0.01$) when compared with age-matched controls.

Table 4. Sensitivity and Specificity of Volume Ratio When Threshold = 0.5*

	T+ (V Ratio >0.5)	T– (V Ratio ≤0.5)
Cases (coronoid hypertrophy)	12	0
Normal controls	1	23

T+, positive test (V or SA >0.5); T–, negative test (V or SA ≤0.5).
*Sensitivity, 100 percent; specificity, 96 percent; positive predictive value, 92 percent; negative predictive value, 100 percent.

Table 5. Sensitivity and Specificity of Surface Area Ratio When Threshold = 0.5*

	T+ (SA Ratio >0.5)	T– (V Ratio ≤0.5)
Cases (coronoid hypertrophy)	12	0
Normal controls	2	22

T+, positive test (V or SA >0.5); T–, negative test (V or SA ≤0.5).
*Sensitivity, 100 percent; specificity, 92 percent; positive predictive value, 86 percent; negative predictive value, 100 percent.

and 86 percent, respectively. The negative predictive values were 100 percent for both tests.

DISCUSSION

Establishing a reliable method for quantitatively studying the mandible is important because, although coronoid hypertrophy seems intuitive to identify subjectively, it is nevertheless difficult to define and measure *objectively* (i.e., how large is a hypertrophied coronoid?). Many studies cite the need for adequate radiographic assessment to properly assess coronoid morphology, the relationship of the coronoid and the zygoma, and temporomandibular joint pathology that may contribute to coronoid hypertrophy.[2,7,8] To date, however, there has been no consensus on how to quantify the volume and surface area of the coronoid process.

In this study, we developed a method to quantitatively measure coronoid and condylar size based on three-dimensional medial axis analysis.[6] This method represents a divergence from traditional two-dimensional cephalometric techniques, in that it allows for accurate and reproducible isolation of the coronoid process and condyle without relying on extramandibular structures or arbitrary fixed points, which may be altered in pathological conditions.[6] By utilizing three-dimensional medial axis analysis to describe coronoid and condylar geometry, our method is broadly applicable across a wide range of coronoid shapes and sizes and is not dependent on a consistent orientation of the mandible (e.g., in full occlusion) at the time of imaging.

Our data demonstrate that coronoid hypertrophy can be defined by volume and/or surface area ratios. Relative size comparison to the ipsilateral condyle is essential since the *absolute* bony volume or surface area measurements vary considerably with patient age, sex, and pathology. In contrast, the relative volume and surface area *ratios* demonstrated small variance and were thus more suitable for comparative analysis. Specifically, significant coronoid:condyle disproportion was evidenced by a volume ratio greater than or equal to 0.5 (sensitivity, 100 percent; specificity, 96 percent) or surface area ratio greater than or equal to 0.5 (sensitivity, 100 percent; specificity, 92 percent). The most severe cases of coronoid hypertrophy featured volume and surface area ratios greater than or equal to 1.0 (cf. Table 3; cases 1, 5, 7, and 12).

These threshold values may serve as a basis for objectively defining coronoid:condyle disproportion, grading severity, guiding treatment, and monitoring for postoperative improvement or recurrence.[2] In our patient population, coronoid: condylar disproportion was due exclusively to cases of coronoid hypertrophy; however, it is necessary to note that an elevated coronoid:condyle volume or surface area ratio may also exist in the case of the normal coronoid but underdeveloped condyle. In this situation, the better treatment might not consist of coronoidectomy but rather condylar reconstruction by grafting or distraction osteogenesis. Thus, despite the utility of our metric in identifying coronoid:condylar disproportion, clinical judgment is always necessary to discern the best course of management.

We believe that three-dimensional medial axis analysis is a useful adjunct for the objective diagnosis of coronoid hypertrophy and is easily implemented using current standard craniofacial computer tomographic data. Longitudinal studies are underway to correlate specific values (i.e., a ratio of 0.5 versus 1.0) with the natural temporal progression of the disorder and, specifically, with functional deficits, such as decreasing interincisal opening, that warrant operative intervention.

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