Cone beam computed tomography signs of desmoplastic ameloblastoma: review of 7 cases

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Objective. This study aimed to evaluate and summarize the radiographic features of desmoplastic ameloblastoma (DA) on cone beam computed tomography (CBCT) scans and to provide necessary further information for clinical diagnosis and preoperative assessment.

Study Design. Seven cases with pathologic diagnosis of DA were studied retrospectively. A CBCT scan of each individual was analyzed and compared with its corresponding plain (panoramic) radiograph.

Results. We found that 71.4% of lesions were located in the anterior or premolar regions (or both) and showed root displacement. With the advent of CBCT imaging, most DA lesions (57.1%) were perceived as having honeycomb appearance totally or dominantly. Distinctively, all the lesions presented cortical expansion with perforation in the buccal/labial side.

Conclusions. The typical intrallesional structure with honeycomb appearance and the dominant buccal/labial cortical expansion with perforation could be proposed as the characteristic features of DA on CBCT images. CBCT can provide more information for preoperative radiologic assessment of DA compared with panoramic radiography. (Oral Surg Oral Med Oral Pathol Oral Radiol 2014;118:e126-e133)

Desmoplastic ameloblastoma (DA) is a rare variant of intraosseous ameloblastoma (IA). It was initially reported in 1984 and termed “ameloblastoma with pronounced desmoplasia” by Eversole et al. In 2005, the World Health Organization classification of odontogenic tumors adopted DA as a histologic type of ameloblastoma with distinct clinical, radiologic, and pathologic features.

DA has a predilection for the anterior or premolar region in the mandible or maxilla. On plain radiographs, this tumor frequently presented as a mixed radiolucent/radiopaque lesion with diffused surrounding features, whereas the majority of other IA variants are predominantly radiolucent. Therefore, it was difficult to differentiate DA from fibro-osseous lesions in half of the DA cases. Histologically, this variant was characterized as extensive stromal collagenization or desmoplasia with small nests and strands of odontogenic epithelium inside.

Accurate radiologic examination of the lesion and its relationship with surrounding anatomic structures is essential preoperatively. Compared with plain radiographic images, cone beam computed tomography (CBCT) scans can provide 3-dimensional (3D) information of the imaged area and more accurately visualize the internal structure and expansion of maxilla-mandible tumors, which can facilitate optimal preoperative planning. However, no previously published articles have focused specifically on the features of DA in CBCT images. By comparing the DA appearances on CBCT and panoramic images, the present study aimed to summarize the features of DA on CBCT images and to provide necessary further information for clinical diagnosis and preoperative assessment.

MATERIALS AND METHODS

Participant recruitment

Patients registered in the West China Stomatology Hospital, Sichuan University, China, with postoperative pathologic diagnosis of DA from May 2011 to April 2013 were recruited in this retrospective study. Postoperative recurrent cases were not eligible to be included in this study.

Before any invasive procedure was performed, each patient had gone through panoramic and CBCT imaging in the Department of Oral Radiology. Two radiologists separately evaluated the radiologic features of each

Statement of Clinical Relevance

Cone beam computed tomography can be more helpful (compared with panoramic radiography) in showing the border, internal structure, cortical expansion with erosion, and surrounding structures, which provide radiologists and clinicians more information for preoperative diagnosis of desmoplastic ameloblastoma, such as the honeycomb appearance and labial/buccal expansion with erosion.
patient, and the preoperative diagnosis was determined by 1 clinician together with the 2 radiologists. The pathologic diagnosis was achieved by means of postoperative biopsy and confirmed by 2 pathologists. Clinical data were obtained from the database of patient records in the Department of Maxillofacial Surgery.

Data collection
Clinical data including age at time of diagnosis, gender, location (mandible or maxilla), and initial treatment were collected. Radiologic data of the DA lesion on both panoramic and CBCT images were recorded, including site (presented by corresponding tooth position), locularity (unilocular, multilocular, or honeycomb), radiodensity (radiolucent, mixed, or hybrid), borders (well or ill defined), and effect on involved teeth (impaction, displacement, or resorption). Some other 3D features, including expansion of the jaw (with the change of cortex) and effects on surrounding structures such as maxillary sinus or mandibular canal, were evaluated only on CBCT images.

As for the tooth position, incisors and canines were regarded as the anterior region. The premolar region is from the first premolar to the second premolar. The molar region is from the first molar to the ramus (mandible) or tuberosity (maxilla). On a panoramic image, a lesion was considered as unilocular when it presented as a single mixed radiolucent/radiopaque mass, whereas a lesion was perceived as multilocular when it was divided into various compartments by septa. In comparison, a lesion on a CBCT image was considered as unilocular when a single cystic cavity was present; as multilocular when septa divided the cyst to be soap-bubble shape; or as honeycomb when septa divided the lesion into a honeycomb shape. The honeycomb shape included cases that contained a large cyst hybridized with the honeycomb portion.

The DA lesions in our study were considered as radiolucent when the shade was presented as a radiolucent cyst; as mixed when it was radiolucent/radiopaque; or as hybrid when the lesion contained both mixed structure and radiolucent cyst. The border was evaluated as well defined when the lesion was clearly demarcated or as ill-defined when the lesion had an indistinct boundary. The mandibular canal and maxillary sinus were defined as being affected when their anatomic structures were changed by the lesions.

Panoramic and CBCT projection
The panoramic radiographs were obtained using the Orthopantomograph OP200 D (Instrumentarium Dental). The device was operated at 8 mA, 60 kV with a typical exposure time of 14.1 seconds. All CBCT examinations were performed using the 3D Accuitomo 200 (J. Morita Mfg Corp, Kyoto, Japan). The parameters were set at 4 mA, 85 kV, and a typical exposure time of 21 seconds, circling the head in 1 rotation.

RESULTS
Among the 7 cases, the male-to-female ratio was 1.3:1. The age ranged from 30 to 57 years, with a peak (57.1%) in the fourth decade of life and a mean age of 42 ± 8 years. All 7 cases were intraosseous lesions (42.9% [3 of 7] occurred in the maxilla, and 57.1% [4 of 7] occurred in the mandible); 85.7% of patients (6 of 7) were treated with resection, whereas only 14.3% (1 of 7) were treated with curettage. Table I shows the detailed clinical features of all cases.

The common radiologic features in panoramic radiographs and CBCT scans are listed in Table II; 71.4% of lesions (5 of 7) were located in the anterior or premolar regions (or both), and 28.6% of lesions (2 of 7) occurred in the premolar to molar region. Root resorption was found in 28.6% of patients (2 of 7), and root displacement was found in 71.4% (5 of 7). An impacted tooth was found in 28.6% (2 of 7).

On panoramic images, 71.4% of lesions (5 of 7) presented as unilocular and mixed content with

<table>
<thead>
<tr>
<th>Case</th>
<th>Age (y)/Gender</th>
<th>Location</th>
<th>Physical examination</th>
<th>Treatment</th>
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<td>Resection</td>
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<tr>
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<td>MX</td>
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<td>B</td>
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<table>
<thead>
<tr>
<th>Case</th>
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<tr>
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<td>(-)</td>
<td>(-)</td>
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<tr>
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<td>Lower left 2nd premolar - left ramus</td>
<td>(-)</td>
<td>(+)</td>
<td>(+)</td>
</tr>
<tr>
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<td>(-)</td>
<td>(-)</td>
</tr>
<tr>
<td>4</td>
<td>Lower right lateral incisor - lower right 1st molar</td>
<td>(+)</td>
<td>(-)</td>
<td>(-)</td>
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<td>Upper left 1st premolar - upper left 2nd molar</td>
<td>(+)</td>
<td>(-)</td>
<td>(+)</td>
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<tr>
<td>6</td>
<td>Upper left central incisor - upper left 1st molar</td>
<td>(-)</td>
<td>(-)</td>
<td>(-)</td>
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<tr>
<td>7</td>
<td>Lower left 2nd premolar - lower right 1st molar</td>
<td>(+)</td>
<td>(+)</td>
<td>(-)</td>
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</table>

(+), positive results; (-), negative results.
ill-defined border (Figures 1 to 3), whereas the other 28.6% (2 of 7) involved multilocular lesions with well-defined borders, including 14.3% (1 of 7) of radiolucent appearance (Figure 4) and another 14.3% (1 of 7) of hybrid appearance (Figure 5). All the features are described in detail in Table III.

As for the appearances of DA on CBCT images, the detailed features are listed in Table IV. All the lesions exhibited well-defined borders on CBCT images; 57.1% (4 of 7) were of the honeycomb type, in which half of lesions showed honeycomb-like mixed content (see Figure 1) and the other half were hybrids of honeycomb-like structure combined with a cystic portion (see Figure 2). Whereas 28.6% of the lesions (2 of 7) had unilocular appearance with mixed shade (see Figure 3), 14.3% (1 of 7) displayed radiolucent multilocular appearance (see Figure 4).

With regard to the radiologic expansion on CBCT, 85.7% (6 of 7) exhibited buccolingual or labial-palatal expansion, which mainly involved the labial/buccal expansion with thinning of the lingual/palatal cortex, whereas 14.3% (1 of 7) exhibited only buccal expansion. All lesions showed cortical erosion in labial/buccal areas. Mandibular canals were displaced in 42.9% of cases (3 of 7), and 28.6% of cases (2 of 7) showed deformation of the sinus cavity.

**DISCUSSION**

DA is a rare variant of ameloblastoma, with an incidence of 0.9% to 13% among all ameloblastoma cases...
in the literature.\textsuperscript{3,4,6,7} It is considered as having a pre-
dilection for the anterior and premolar regions of the
jaw, and tooth displacement frequently happens in
those regions.\textsuperscript{3-6} One author team summarized that their
patients with DA had an average age of 42.9 years, a
male-to-female ratio of approximately 1:1, and a
maxilla-to-mandible ratio of 1:0.9.\textsuperscript{7} In our study, the
distributions of age, gender, and location were found to
be consistent with those numbers.

Previous studies of DA have summarized features of
most lesions on plain radiography as mixed radiolucent/
radiopaque appearance with ill-defined borders; thus,
they resembled fibro-osseous lesions.\textsuperscript{5-7} In agreement
with these studies, the appearance on panoramic radi-
ography of DA cases in our study showed a similar
pattern. Therefore, it appeared that panoramic radiog-
raphy cannot provide adequate information to distin-
guish DA from fibro-osseous lesions.

Panoramic radiography has inevitable disadvantages
of geometric distortion, superimposition of anatomic
structures, and poor capability for showing fine
details.\textsuperscript{9,10} The anterior or premolar regions, in which
DA usually occurs, were found to have more overlap
with adjacent structures and higher distortion on
panoramic images. For instance, DA located in the anterior maxilla region was commonly superimposed with the nasal basis and maxillary sinus. In this regard, panoramic radiography has intrinsic limitations on its ability to present the fine mixed internal structure of DA.

Compared with panoramic radiography, the 3D view of CBCT can display the detailed internal structure of DA. As a newer CT modality, CBCT has a relatively higher isotropic spatial resolution of osseous structures at lower doses of radiation and lower financial cost than multidetector CT.9,11

In the previous literature, the lesions with mixed radiolucent/radiopaque content were variously described as “granular or cloudy,” “needle-like trabecular,” “highly dense trabecular,” or having “honeycomb appearance.”6,12 In our study, a lesion with honeycomb structure was the dominant type of DA. We further classified the lesions with a honeycomb-like area conjugated with a radiolucent cystic portion as the hybrid

Fig. 3. A. Panoramic view shows a unilocular mixed lesion with an ill-defined border in the left maxilla. B-D, Cone beam computed tomography 3-dimensional images indicate a well-defined cystic lesion within flecks of calcification (arrow) and show that the left canine is impacted. The floor of the left maxillary sinus was pushed up. E, Histopathologic image shows a few epithelial islands (arrow) scattered in extensive desmoplasia with metaplastic bone (original magnification ×40).
type, in which the radiolucent cyst varies in its size and site in individual cases. In some special cases, the hybrid type may be concealed on panoramic radiography when the radiolucent cyst is located at the labial or lingual side of the lesion and completely overlaps with the honeycomb-like area.

Through the comprehensive analysis of DA images, we suggest that a characteristic CBCT feature of DA is the honeycomb-like appearance formed by coarse trabecular septa, which was found in more than half of our cases. It is obviously distinct from the typical radiographic features of other types of ameloblastoma, which are often perceived as unilocular or multilocular radiolucent cystic lesions. With clear observation of internal structures in tumors, CBCT can also better distinguish DA from fibro-osseous lesions compared with panoramic radiography. Besides the honeycomb-like features, some DA lesions have plenty of radiopaque flecks scattered around the radiolucent region, which is in agreement with the findings of other authors. This feature differs from the unicystic type of ameloblastoma, which often shows a radiolucent cystic lesion without a radiopaque area.

Fig. 4. A, Panoramic view shows a multilocular radiolucent lesion with a well-defined border and root resorption (arrow) in the left mandible. B-D, Cone beam computed tomography 3-dimensional images indicate a multilocular radiolucent lesion with soap-bubble appearance (arrow) and buccal expansion with cortical perforation. E, Histopathologic image shows very irregular tumor islands embedded in stroma with massive desmoplasia, without any metaplastic bone (original magnification ×40).
Another characteristic CBCT feature of DA that we have proposed is the apparent expansion of a lesion in the labial/buccal side with partial cortical erosion, whereas other common IA lesions often exhibit buccolingual expansion with perforation.\textsuperscript{14,15} According to Sheikh et al.,\textsuperscript{4} this feature can adequately confirm that a DA lesion has potentially aggressive behavior. The fact that most cases in this study were treated with partial resection of the jaw also reflects the concern about this behavior of DA. Although approximately half of reported DA lesions had a diffuse radiologic border, which may indicate that this tumor is more aggressive than other ameloblastoma variants,\textsuperscript{6,16} all cases in our study had well-defined borders on CBCT images. It appears that the 3D images can

Table III. Panoramic presentation of 7 cases of desmoplastic ameloblastoma

<table>
<thead>
<tr>
<th>Case</th>
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<th>Border</th>
<th>Radiologic content</th>
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<td>7</td>
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\textsuperscript{U}, unilocular; \textsuperscript{M}, multilocular; \textsuperscript{I}, ill-defined; \textsuperscript{W}, well-defined; \textsuperscript{MI}, mixed; \textsuperscript{RA}, radiolucent; \textsuperscript{HY}, hybrid.

Another characteristic CBCT feature of DA that we have proposed is the apparent expansion of a lesion in the labial/buccal side with partial cortical erosion, whereas other common IA lesions often exhibit buccolingual expansion with perforation.\textsuperscript{14,15} According to Sheikh et al.,\textsuperscript{4} this feature can adequately confirm that a DA lesion has potentially aggressive behavior. The fact that most cases in this study were treated with partial resection of the jaw also reflects the concern about this behavior of DA. Although approximately half of reported DA lesions had a diffuse radiologic border, which may indicate that this tumor is more aggressive than other ameloblastoma variants,\textsuperscript{6,16} all cases in our study had well-defined borders on CBCT images. It appears that the 3D images can

Fig. 5. A, Panoramic view shows a hybrid lesion comprising a mixed area combined with large cystic changes (mesial-distal) in the anterior-premolar region of the mandible. Root displacement (black arrow) and root resorption (white arrow) were seen. B-D, Cone beam computed tomography 3-dimensional images indicate a hybrid lesion with expansion and cortical perforation labially. The large cystic changes were mesial-distal and labial (arrow). E, Histopathologic images exhibit the extensive collagenized stroma containing tumor epithelial islands with metaplastic bone (original magnification $\times40$). Cystic degenerations were seen in the epithelial islands (arrow) (original magnification $\times100$).
avoid the tissue superimposition and provide a more accurate and clearer border.

By analyzing the radiographic and histopathologic images, we found that the metaplastic bone in desmoplastic stroma was observed only in DA lesions with mixed shade. However, there is no agreement on whether the mixed radiolucent/radiopaque structures came from new bone formation or residual bone of tumor destruction.6-8,16 In addition, microcystic degeneration was observed in the epithelial islands in the hybrid lesions in our cases. It was speculated that the radiolucent cyst in a hybrid lesion might be developed from these microcysts in the solid mass of DA. This finding corresponded with some previous studies.7,17

CONCLUSION
CBCT is advantageous in perceiving the borders, internal structure, cortical expansion, and relationship with surrounding structures of DA. A typical intralobular structure of honeycomb appearance and the dominant labial/buccal expansion with cortical erosion was observed in the epithelial islands in the hybrid lesions in our cases. It was speculated that the radiolucent cyst in a hybrid lesion might be developed from these microcysts in the solid mass of DA. This finding corresponded with some previous studies.7,17

REFERENCES

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