RESEARCH ARTICLE

Characteristic features of the odontogenic myxoma on cone beam computed tomography

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Objectives: To illustrate the characteristic features of odontogenic myxoma (OM) on CBCT.

Methods: From 52 subjects with histopathologically diagnosed OM, 18 subjects who underwent a CBCT examination were retrieved between May 2009 and April 2016. Features on CBCT images and clinical records were carefully observed and analyzed.

Results: Characteristic features include: (1) fine and straight septa that were recognized to separate the tumour into triangular, square or rectangular spaces, which appeared as “tennis racket” or “honeycomb” patterns; (2) septa that frequently scattered to the borders of lesions and appeared perpendicular to the margins; (3) tooth displacement and resorption that were seen in most of the OM lesions; (4) OM that were noted to have a tendency to involve the alveolar process, scallop between the roots and affect the integrity of the alveolar ridge; (5) the cortex of OM that appeared normally perforated and the edge of the cortex expanded into the soft tissue.

Conclusions: CBCT is highly effective in demonstrating the comprehensive internal structures of the lesions precisely and providing detailed information for the diagnosis of OM. *Dentomaxillofacial Radiology* (2017) **46**, 20160232. doi: [10.1259/dmfr.20160232](https://doi.org/10.1259/dmfr.20160232)

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Introduction

Odontogenic myxoma (OM) is an odontogenic neoplasm, accounting for 3–6% of all odontogenic tumours.^{1,2} OM of the jaws was first described by Thomas and Goldman in 1947.³ Its pathologic feature is an accumulation of mucoid ground substance with little collagen. The amount of collagen determines whether it can be called a myxofibroma.^{4,5} According to the World Health Organization classification, OM is a benign tumour of ectomesenchymal origin with or without odontogenic epithelium.³ OM is most commonly found in subjects 20–40 years of age and possesses a female

predilection.^{6,7} The premolar and the molar regions of the mandible are the sites of most common occurrence for OM.^{3,6,8} Patients afflicted with an OM generally notice a painless, slowly enlarging expansion of the jaw with possible tooth loosening or displacement.^{3,4,9} As the tumour expands, it frequently infiltrates adjacent structures. Maxillary lesions often extend into the sinuses, while mandibular tumours often infiltrate the mandibular canal, which makes resection difficult and imposes high chances of recurrence.^{3,10} Therefore, in order to establish a proper diagnosis and provide detailed information regarding the tumour extent, border and involvement with surrounding structures, the use of radiographs is required as a diagnostic and pre-surgical standard.

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Figure 1 Distribution of 18 odontogenic myxomas by age and gender.

The features of OM on conventional radiographs have been well documented in the literature. Most lesions of OM are separated by straight septa forming square, rectangular or triangular spaces and show a “honeycomb”, “soap bubble” or “tennis racquet” appearance.^{3,6,8,11} However, plain radiography is not reliable enough to demonstrate the extent and fine inner structures of the tumour.^{3,12} Therefore, the use of advanced medical imaging including MRI, CT and CBCT are useful in the aid of establishing a proper diagnosis with the ability of clearly displaying a tumour in three-dimensional accuracy and perspective.^{3,13}

CBCT can achieve a high spatial resolution and display more characteristic radiographic findings.^{14,15} It is suitable for the radiographic diagnosis of jawbone lesions and odontogenic tumours with hard-tissue components.^{2,15,16} However, few descriptions of the CBCT features of OM have been published. This study described the clinical and radiographic features of 18 OM subjects in the West China Hospital of Stomatology over the past 5 years. Its aim was to illustrate the characteristics of OM on CBCT images and its differential diagnosis.

Materials and methods

18 subjects were selected from 52 OM subjects histopathologically confirmed by the Department of Oral Pathology in the West China Hospital of Stomatology (Chengdu, China) during May 2009–April 2016. Each

patient had gone through CBCT imaging in the Department of Oral Radiology. All the CBCT images were performed with 3D Accuitomo (J Morita Mfg. Corp., Kyoto, Japan) in its standard mode. The radiation dose (75–78 kV; 2–3 mA) was optimized properly, and radiation protection was performed regularly. Two radiologists separately evaluated the radiologic features of each patient, and the pre-operative diagnosis was determined by one clinician together with the two radiologists. The pathologic diagnoses were achieved by post-operative biopsy and confirmed by two pathologists. Clinical data were obtained from the database of patient files in the Department of Maxillofacial Surgery.

Generally, the clinical data collected included the age and gender of the patient, onset and duration of disease, chief complaint and site of the disease (presented by corresponding tooth position). Radiological data of the OM on CBCT images were recorded and evaluated, including their locularity (unilocular or multilocular), radiodensity (radiolucent, radiopaque or mixed), borders (well or ill defined), margins (clear or unclear), the size of lesions in three dimensions and effects on the surrounding structures including the teeth involved, maxillary sinus or mandibular canal and local alveolar bone. As for the tooth position, incisors and canines were designated as the anterior region, and the region from the first premolar to the ramus (mandible) or tuberosity (maxilla) was designated as the posterior region. Multilocular OMs were classified into “tennis racquet” or “honeycomb” types according to their inner structures. The septa of honeycomb type were thicker with smaller compartments, while the septa of the tennis racquet type were thinner with relatively larger compartments. The lesions were measured in millimetres along their longest axis, getting the value of anteroposterior and buccal–lingual in horizontal position, the value of superoinferior in sagittal position. All the measurements were conducted by two radiologists.

Table 1 Location of 18 odontogenic myxomas

Area	Maxilla	Mandible	Total
Anterior area	0	0	0
Posterior area	2	9	11 (61%)
Anterior and posterior area	1	2	3 (17%)
Lesions that cross the midline	3	1	4 (22%)
Total	6 (33%)	12 (67%)	18

Table 2 CBCT appearance of 18 odontogenic myxomas

	Unilocular		Multilocular		Total
	Maxilla	Mandible	Maxilla	Mandible	
Border					
Well defined	1	2	4	7	14
Ill defined	0	0	1	3	4
Margin					
Clear	0	2	0	1	3
Unclear	1	0	5	9	15
Effect on involved teeth					
Displacement only	1	0	3	1	5
Resorption only	0	1	0	0	1
Combination of displacement and resorption	0	0	2	5	7
Neither displacement nor resorption	0	1	0	4	5
Involved alveolar					
With alveolar ridge continuity	0	0	1	4	5
Without alveolar ridge continuity	1	0	4	5	10
Uninvolved alveolar	0	2	0	1	3

Results

The distribution of OMs by age and gender is shown in Figure 1. The age range was from 6 to 75 years with a mean age of 35.5 years, and 60% subjects were diagnosed between the second and fifth decades of age. The male-to-female ratio is 1:2. The duration of the disease ranges from 2 months to 10 years. 12 subjects had complaints of a slowly growing and painless lesion with facial asymmetry, while 3 cases were identified through medical examination. Only three patients presented with a complaint of pain, one of which was probably due to the local infection. The location of the lesions is given in Table 1. Six of the lesions that occurred in the mandible involved the mandibular ramus, while one lesion located in the chin extended bilaterally from the right canine to the left first premolar.

The radiographic features of CBCT scans are noted in Table 2. 15 multilocular lesions were observed. 10 (67%) lesions were located in the mandible and 5 lesions in the maxilla. One case showed a honeycomb pattern (Figure 2), nine subjects showed a tennis racquet pattern (Figure 3) and three subjects showed a mixed appearance of honeycomb and tennis racquet patterns. The rest of the five subjects showed fewer septa than the

above subjects (Figure 4). Six subjects had fine straight septa perpendicular to the margin (Figures 3 and 4). 3 (60%) out of 5 maxillary myxomas involved the maxillary sinus and nasal cavity (Figure 5). One case of the maxillary myxomas had spread bilaterally involving the maxillary sinus, nasal cavity, ethmoid, hard palate, zygomatic bone and orbital floor. One lesion appeared to displace the floor of the maxillary sinus upwards (Figure 3). Five mandibular multilocular lesions displaced the mandibular canal. One case perforated the superior wall of the mandibular canal (Figure 4). All 15 multilocular lesions showed mixed density and perforated cortex, while only 8 (53%) lesions showed that the cortex had intruded in the soft tissues (Figure 6). Two cases showed a “sunburst” appearance, indicating that the cortex was perforated and the radiopaque lines extended from the periosteum (Figures 3, 6).

Three OMs were unilocular, presenting with two lesions in the posterior of the mandible and one lesion in the posterior of the maxilla. One lesion was located between the roots of two teeth and another involved the dental follicle of the impacted third molar in the left mandible. The above two subjects showed perforation of the cortex. The remaining lesion surrounded the

Figure 2 The multilocular lesion that occurred in the right posterior of the mandible: the axial view (a) is showing a lesion with an ill-defined border and diffuse margin. The sagittal view (b) is showing that the septa are thick, resembling a honeycomb pattern (arrow). The coronal view (c) is showing that the cortex is perforated.

Figure 3 The axial view (a) and sagittal view (b) of the left maxilla are showing that the septa (white arrow) have scattered in the periphery of the lesion showing the pattern of the strings of a tennis racket. The sagittal view (c) and coronal view (d) are showing a sunburst appearance (black arrows) and that the lesion has involved the left maxillary sinus and the alveolar process.

crown of the impacted premolars in the right mandible with the cortical bone intact (Figure 7). All the three lesions appeared radiolucent.

The size of the OMs is shown in Figure 8. The size of unilocular lesions ranged from 3 to 14 mm (mean 7.0 ± 5.0 mm) in the anteroposterior direction, from 3 to 14 mm (mean 9.7 ± 2.9 mm) in the buccal–lingual direction and from 9.0 to 12 mm (mean 10.3 ± 1.2 mm) in the superoinferior direction. Whereas for multilocular lesions, the size ranged from 10 to 69 mm (mean 37 ± 17.0 mm) in the anteroposterior direction, from 9 to 44 mm (mean 21.5 ± 10.3 mm) in the buccal–lingual direction and from 11 to 58 mm (mean 32.4 ± 15.1 mm) in the superoinferior direction.

Discussion

Previous literature of OM have summarized its features on plain radiography and CT. Compared with plain radiography, CBCT can avoid geometric distortion, superimposition of anatomic structures and display the fine internal structure of the lesions precisely.^{2,16} There are very few reports with such a large number of subjects in the literature focused on features of OM on CBCT images. In this study, we analyzed the clinical and radiographic features of 18 OM subjects and

illustrated the characteristic features of OM on CBCT in detail.

This study presented a wide age range (6–75 years), in which few subjects were diagnosed in the first or sixth decades. There was a female predilection with a male-to-female ratio of 1:2. The chief complaint of the patients was a painless swelling in the affected area with subsequent facial asymmetry. Only three patients complained of pain and one of them might have been due to infection. In terms of tumour site, both unilocular and multilocular lesions were located most frequently in the posterior region of the jaw in this sample. The findings above correspond to previous studies on OMs.^{3,6,8}

Histologically, the trabeculae of woven bone were seen dispersing in 25.8–56% lesions. Radiologically, these residual bony trabeculae and dense fibrous tissue manifested as septa in multilocular OMs.⁸ As reported previously, fine and straight septa separated the lesion into triangular, square or rectangular compartments, which are described as “honeycomb” or “tennis racket”.^{3,6,8,11} These features significantly aided in the proper diagnosis of OM. In this present study, only 10 multilocular OMs showed that typical appearance, and the remaining 5 demonstrated fewer septa. Most septa were distributed at the periphery of the lesions, which concurred with previous literature.¹¹ Furthermore, we found that the septa were perpendicular to the margins

Figure 4 The axial view (a), sagittal view (b) and coronal view (c) are showing that the tumour located in the right posterior region of the mandible and the continuity of the cortex has been interrupted, and the edge of the cortex (white arrows) has expanded into the soft tissue lingually. The sagittal view (b) is showing that the septa are perpendicular to the margins (black arrow).

Figure 5 CBCT showing the tumour located in the left side of the maxilla: the axial view (a) is showing that the tumour has involved the maxillary sinus, hard palate and nasal cavity. The sagittal view (b) is showing that the second and third molars are displaced. The coronal view (c) is showing that the mesiobuccal root of the first molar has been absorbed.

in six lesions. In comparison, the septa of other odontogenic tumours such as ameloblastoma and keratocystic odontogenic tumour were not perpendicular to the marginal cortex. We assume that the perpendicular septa benefit the diagnosis of OM. All multilocular OMs showed mixed density because of the septa inside, while all unilocular OMs were radiolucent.

OMs appear histologically as an abundant myxoid or mucoid extracellular matrix, and mostly without capsule.^{4,10,17} These characters explain the aggressive nature of the pathology. Although the borders of 14 (78%) subjects were well defined, 15 (83%) subjects showed an unclear margin in our study. All multilocular lesions and two unilocular lesions showed that the lesions interrupted the cortical continuity, and the cortical edge of eight lesions intruded into the soft tissue. Two lesions showed radiopaque lines with a perpendicular orientation extending from the perforated cortex into the soft tissue forming a “sunburst” appearance. Some studies reported that these features suggest the invasive nature of the tumour radiographically and advised differentiating from metastatic lesions in the jaw and osteogenic sarcoma.^{3,6,11,12} Koseki *et al*¹⁰ reported that OMs tended to involve the alveolar process in tooth-bearing areas. 5 (33%) of 15 multilocular subjects scalloped between the roots and 9 (60%) subjects interrupted the integrity of alveolar ridge in this

study. Tooth displacement (79%) is seen more often than tooth resorption (43%), both of which were higher than 69% and 3.8% reported by Titinchi *et al*.⁸ 3 (60%) of the 5 maxillary multilocular lesions showed encroachment on the maxillary sinus and nasal cavity, with 1 lesion extensively affecting the maxillary sinus, nasal cavity, hard palate, zygomatic bone and orbital bone. 4 (80%) of 5 subjects located in the posterior of the mandible compressed the mandibular canal instead of encroaching. These findings are consistent with previous literature.^{8,10} As for the size of the lesions, the mean size of multilocular lesions was significantly larger than that of unilocular lesions in all three dimensions. This result was consistent with the report on plain radiography by Titinchi *et al*.⁸

The radiographic appearance of one unilocular OM showed atypical characteristics. This lesion occurred in the posterior of the mandible and surrounded the crown of the impacted premolars in the right mandible. It showed a well-defined border, clear margin, homogeneous density and cortical continuity. As a result, the inclination is to make a diagnosis of dental papilla of a developing tooth rather than OM according to its radiological features, although the pathologists prefer to diagnose the lesion as OM because of its myxoid change under microscope. In reference to the literature, the histopathology of OM needs to be differentiated from

Figure 6 CBCT showing the tumour located in the anterior of the mandible: the axial view (a), sagittal view (b, c) and coronal view (d) are showing that the cortex is perforated and radiopaque lines (arrows) are extending from the periosteum, resembling a “sunburst” appearance. The alveolar process is involved.

Figure 7 The axial view (a) is showing that the tumour is located in the right mandible. The sagittal view (b) and coronal view (c) are showing that this lesion has surrounded the crown of the impacted premolars with a well-defined border, clear margin, homogeneous density and cortical continuity.

various benign lesions with a myxoid change including the dental papilla of a developing tooth.^{5,18}

OMs have a variable radiographic appearance and can mimic various lesions of the jaw, which often leads to a misdiagnosis. OMs should be differentiated from ameloblastomas, central giant-cell granulomas and intraosseous haemangiomas.^{3,4,8} The compartments of an ameloblastoma are round-like rather than the square or triangular spaces of OMs. Compared with OMs, the margin and the cortical boundary of an ameloblastoma are more defined, and an ameloblastoma is less likely to invade soft tissues or interrupt the cortical continuity.² Luo *et al*¹⁶ reported that desmoplastic ameloblastoma presented as a “honeycomb” appearance, analogous to OMs. However, desmoplastic ameloblastoma commonly involves the anterior and premolar regions of the jaw and has a high tendency to expand at the labial–buccal side.¹⁶ Central giant-cell granuloma frequently presents in patients younger than 30 years of age and occurs in the anterior mandible and incisor region of the maxilla.³ Furthermore, central giant-cell granuloma shows an apparent expansion, which leads to thinning of the cortex. Intraosseous haemangioma usually affects the mandibular canal, causing an abnormal enlargement of the mandibular canal, mental foramen and mandibular foramen. Intraosseous haemangiomas may also present with gingival redness

and haemorrhage. A biopsy is necessary to confirm an accurate diagnosis of OM.^{4,8}

As recommended in previous literature, the treatment of OM depends on the size and behaviour of the lesion and the age of the patient.^{9,17,19,20} A radical therapy is essential when a locally aggressive behaviour is encountered owing to the high rate of recurrence.¹⁷ If the patient is young, a more conservative procedure is suggested because of tumour-related complications.^{19,21} In this sample, all the patients adopted radical treatments and thus far have shown no recurrence.

CBCT was effective in demonstrating the inner structure of OMs. Owing to the high rate of recurrence, the accurate radiographic appearance was critically important in arriving at a correct diagnosis and in surgical planning. In conclusion, the characteristic features of OM include the following points: (1) fine and straight septa separating the lesion into triangular, square or rectangular spaces, which appear as “tennis racket” or “honeycomb” patterns; (2) septa that frequently scattered to the borders of lesions and appeared perpendicular to the margins; (3) tooth displacement and resorption that were seen in most of OM lesions; (4) OMs that were noted to have the tendency to involve the alveolar process, scallop between the roots and affect the integrity of the alveolar ridge; (5) the cortex of OMs that appeared normally perforated and the edge of the cortex expanded into the soft tissue.

Figure 8 The size of odontogenic myxomas: the first 3 are unilocular and the following 15 are multilocular.

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