Radiographic features of a patient with both cemento-ossifying fibroma and keratocystic odontogenic tumor in the mandible: a case report and review of literature

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We herein describe a rare case of a 48-year-old woman with both ossifying fibroma (OF) and keratocystic odontogenic tumor (KCOT) in the mandible. CT images showed a 15 × 15 × 20-mm radiolucent-radiopaque lesion with bucco-lingual bony expansion in the left first premolar equivalent area of the mandible, and a 15 × 40 × 35-mm well-defined unilocular radiolucent lesion in the left side of the mandible, extending from the distal side of the distal root of the left second molar to the left mandibular ramus.

A biopsy of the radiolucent-radiopaque lesion and fenestration surgery of the radiolucent lesion were performed. Histopathologic examination revealed a fibro-osseous lesion (FOL) and a KCOT, respectively.

CT was useful in diagnosing the radiolucent-radiopaque lesion as OF and for detecting the 3-dimensional bone expansion and the contents in the lumen of the KCOT. **(Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;112:798-802)**

Ossifying fibroma (OF) was defined in 2005 by the World Health Organization (WHO) as "a well-demarcated lesion composed of fibrocellular tissue and mineralized material of varying appearances."[1] OF is a benign odontogenic fibro-osseous tumor with a slowly progressing enlargement arising from the periodontal ligament, and is largely restricted to the tooth-bearing areas of the jaws.[2] The popular belief is that the highest incidence occurs during the third and fourth decades of life, with a predilection for women, and most often occurs in the mandibular premolar-molar region.[2] Occasionally, it may grow to massive size, causing considerable cosmetic and functional deformity.[3] Complete surgical removal is necessary to prevent recurrence.[3]

Keratocystic odontogenic tumor (KCOT), formerly known as odontogenic keratocyst (OKC), received its new designation by WHO in 2005 to better convey its neoplastic nature.[1] KCOT develops from the dental lamina, which is found throughout the jaw and overlying alveolar mucosa, and is lined by stratified keratinizing squamous epithelium.[4] The frequency of OKC has been reported to be between 4% and 12% of all odontogenic cysts.[5] The lesion usually occurs in the second and the third decades of life, with a slight male predilection.[6] The lesion is most commonly located in the body or ramus of the mandible.[4] Although approximately half of patients are asymptomatic, pain, swelling, expansion, drainage, and bone perforation have been reported.[7] Generally, KCOT is a solitary lesion unless the lesion is associated with basal cell nevus syndrome (Gorlin-Goltz syndrome).[4] KCOT has been gaining particular interest because of its specific histopathologic features, high recurrence rate, and aggressive behavior.[7] The frequency of recurrence after incomplete surgical treatment has been reported to vary from 10% to 62%.[8]

Although OF and KCOT are relatively common diseases, the cases of patients with both OF and KCOT at

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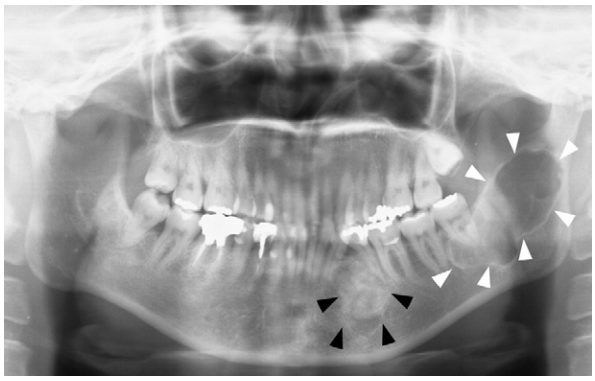

Fig. 1. The panoramic radiograph. Two abnormal lesions in the left side of the mandible, including a well-defined radiolucent-radiopaque lesion (*black arrowheads*) and a well-defined unilocular radiolucent lesion with a cortical margin (*white arrowheads*), can be detected.

different locations in the mandible are very rare. To our knowledge, there has been no previous report of a patient with both OF and KCOT at different locations in the jaws. We herein describe a rare case of a patient with both OF and KCOT at different locations in the mandible, focusing on the radiographic features of the lesions.

CASE REPORT

A 48-year-old woman was referred to our hospital for further investigation and treatment of an asymptomatic radiolucent-radiopaque area and a radiolucent area in the mandible, which had been incidentally detected on a panoramic radiograph in a general dental clinic.

Oral examination demonstrated only a bone expansion of the left premolar equivalent area of the mandible. There was no specified anamnesis or family history.

A panoramic radiograph showed 2 abnormal lesions in the left side of the mandible, including a well-defined radiolucent-radiopaque lesion and a well-defined unilocular radiolucent lesion with a corticated margin (Fig. 1). The radiolucent-radiopaque lesion was located in the left first premolar equivalent area of the mandible, and the teeth adjacent to this lesion showed displacement. Otherwise, the radiolucent area extended from the distal side of the distal root of the left second molar to the left mandibular ramus on the panoramic radiograph. Although the root of the left third molar was involved in the radiolucent lesion, no root resorption or displacement of erupted teeth was noted.

Computed tomography (CT) images were obtained with CT scanners (Light Speed QX/I, General Electric, Milwaukee, WI) at 120 kVp and 150 mA. The field of view was 25 × 25 cm. The matrix size was 512 × 512, the slice thickness was 2.5 mm with no interslice spacing, and the reconstruction slice thickness was 1.25 mm with no interslice spacing. CT images showed a 15 × 15 × 20-mm radiolucent-radiopaque lesion with bucco-lingual bony expansion in the left first premolar equivalent area of the mandible (Fig. 2, *a*). The lesion consisted of central inhomogeneous mineralization and a thin marginal radiolucent area. The average CT number of the central inhomogeneous mineralization area was approximately 1300 to 1400 HU. The lesion did not show any relationship to the adjacent tooth apices or any evidence of root resorption. Moreover, CT images showed a 15 × 40 × 35-mm well-defined unilocular radiolucent lesion in the left side of the mandible, extending from the distal side of the distal root of the left second molar to the left mandibular ramus. In particular, the CT images showed the presence of a bucco-lingual bony expansion and cortical bone thinning in the left mandibular ramus (Fig. 2, *b* and *c*). Although the average CT number of the lesion was 40 HU, the inside of the lesion showed 100 HU in some areas.

On the contrast-enhanced CT images, neither of the lesions showed contrast enhancement. Based on the CT imaging findings, we diagnosed the radiolucent-radiopaque lesion as OF, and the radiolucent lesion as KCOT.

The left third molar was extracted and the radiolucent lesion was biopsied, and KCOT was established. Therefore, the biopsy of the radiolucent-radiopaque lesion and fenestration surgery of the radiolucent lesion were performed. During surgery, it was noted that the radiolucent-radiopaque lesion was filled with a hard tissuelike mass without cystic lesion, and the surrounding thinned cortex was seen. The radiolucent lesion showed a cystic lesion with a thick wall, filled with a large amount of yellow-white substances.

In the histopathological examination, the radiolucent-radiopaque lesion exhibited immature bone or cementumlike calcified materials and relatively cellular fibrous connective tissue (Fig. 3, *a*). The radiolucent lesion exhibited a loose fibrous cystic wall lined by stratified squamous epithelium, which exhibited a parakeratinizing surface and palisaded basal cell layer (Fig. 3, *b*). From these findings, the radiolucent-radiopaque lesion was diagnosed as fibro-osseous lesion (FOL), and the radiolucent lesion was diagnosed as KCOT histopathologically.

Nine months after the surgery, follow-up CT images showed the recurrence of KCOT and surgery was repeated. Two years after the first surgery, significant change of the radiolucent-radiopaque lesion has not been shown.

DISCUSSION

To our knowledge, KCOT or OKC associated with calcifying odontogenic cyst or dentigerous cyst has been documented at different locations of the same patient (Table I).[9,10] In addition, multiple OFs at different locations in the jaws of the same patient and combined histology of OF with aneurysmal bone cyst or giant cell granuloma in one lesion have been reported.[11-13] However, there is no report of OF simultaneously occurring with other conditions at different locations of the same patient. This is the first report of OF and KCOT occurring at different locations in the jaw of the same patient.

The current histopathological classification of maxillofacial FOL includes OF, fibrous dysplasia (FD), and