Cemento-ossifying Fibroma of Mandible: Report of Two Cases

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Abstract
Cemento-ossifying fibroma (COF) is a relatively rare, benign, nonodontogenic tumor of the jaw, a subdivision of fibro-osseous lesions. Age of occurrence is between 20 to 40 years. Female to male ratio is 5:1, with affinity for posterior mandible region. Aim is to evaluate the principal clinical and radiological features of two classical case reports of cemento-ossifying fibroma with discussion on clinical features and radiographic and imaging features. Here we are presenting two classical case reports of cement-ossifying fibroma of the mandible. Case 1 was a 19 years old female reported with painless swelling of molar-premolar region of right mandible with duration of 6 months. Case 2 was a 23-year-old female with an asymptomatic giant swelling of right mandible with duration of 3 years. Both cases were having typical clinical and radiographic features of COF which was later confirmed by histopathology.

Keywords: Cemento-ossifying fibroma, mandible, molar-premolar region, 3-dimensional CT scan.

INTRODUCTION
Cemento-ossifying fibroma (COF) is a bone producing, slow growing, asymptomatic, well-demarcated, benign lesion of the jaw. Typically affects female between 20 to 40 years, predilection for the mandible premolar-molar region, causing a painless swelling but undergoes slow expansile growth. Most probably tumor originates from periodontal membrane, therefore, with double embryonic origin (ectodermic and mesodermic). In fact, connective tissue of the periodontal membrane can contemporarily elaborate both bone and cementum. Bernier hypothesized that the etiopathogenesis of COF in the bone might be caused by an irritant stimulus (such as tooth extraction) which may activate the production of new tissue from the remaining periodontal membrane. The etiopathogenesis of extraosseous COF, where there is no periodontal tissue as suggested by Cakir and Karadayi is from embryonic nests and from ectopic periodontal membrane as suggested by Brademann et al cytogenetic studies are limited.

In one study, 3 orbital COF exhibited balanced translocations with recurring breakpoints at Xq26 and 2q33. Dal cin et al also reported an interstitial deletion on chromosome 2 between q31 and q35-36. Radiographically, they are well-defined unilocular or multilocular lesion with smooth contour. Depending on the stage of maturation, cemento-ossifying fibroma can be radiolucent, mixed variety to completely radiopaque. Histologically, it appear as well-circumscribed, occasionally encapsulated with various amount of bony trabeculae/cementum formation in a fibrous stroma.

CASE REPORTS
Case 1
A 19-year-old female reported to our OPD with the chief complaint of a painless, slow-growing, progressive swelling in right mandible body region. Duration of swelling was 6 months. The medical, dental and family histories were unremarkable. On clinical examination, extraorally a small localized swelling of right mandible body region. Duration of swelling was 6 months. Case 2 was a 23-year-old female with an asymptomatic giant swelling of right mandible with duration of 3 years. Both cases were having typical clinical and radiographic features of COF which was later confirmed by histopathology.

Keywords: Cemento-ossifying fibroma, mandible, molar-premolar region, 3-dimensional CT scan.
Based on clinical features and typical radiographic features, provisional diagnosis of benign fibro-osseous lesion, most probably ossifying fibroma was given. Incisional biopsy was taken and the diagnosis was confirmed by histopathology as cemento-ossifying fibroma. Microscopically it showed both bone and cementum component in a fibrous connective tissue stroma (Fig. 4).

Case 2
A 23-year-old female reported with a painless slow-growing swelling in the body of the right mandible. The swelling had first been noticed 3 years previously. The medical and family history was not significant. Dental history included extraction
of 46, 47 due to dental caries at very young age. On clinical examination, extraorally a large swelling on right side of the mandible extending posteriorly to involve the ramus region with bowing of inferior border, leading to overall facial disfigurement with a size of 14 cm × 8 cm. On palpation, swelling was bony hard and nontender. Lymph nodes were not palpable. Intraorally, the 46, 47 teeth were missing. Adjacent teeth 44, 45 and 48 were not displaced. There was expansion of cortical plates with intact overlying gingival mucosa extending from 45 to 48 tooth region. The overlying mucosa was not smooth on alveolar crest of extracted 46, 47. On palpation, the swelling was bony hard, nontender. 44, 45 and 48 were nontender and they gave vital response on vitality testing. Laboratory investigations were all within normal limits. Based on history and clinical examination a clinical diagnosis of a benign lesion was given.

Radiographically, OPG view revealed a well-defined mixed radiolucent-radiopaque lesion of approximately size of 10 cm × 5 cm extending from angle of right mandible crossing the midline up to 33 tooth region posteroanteriorly and superiorly from alveolus of missing 46, 47 teeth to involve inferior border of mandible on right side, inferiorly. Inferior border of mandible was thinned but intact.

On cross sectional mandible occlusal view, buccal and lingual cortical expansion was extending from 43, 44 tooth region to posteriorly on right side with a flocculent pattern and large tufts of bone formation. Cortical plates were intact.

CT scan (Axial image) revealed heterodense, expansile lesion with well-defined borders in body of right mandible.

CT scan (Coronal image) revealed heterodense, expansile lesion with well-defined borders in body of right mandible with mediolateral extent of the lesion.

Three-dimensional CT scan showed a giant lesion involving the right mandible completely, leading to facial disfigurement. (Figs 5A and B).

Provisional diagnosis of benign fibro-osseous lesion, most probably ossifying fibroma was given based upon clinical features and typical radiographic features. Incisional biopsy was taken and the diagnosis was confirmed by histopathology as cemento-ossifying fibroma. Microscopically it showed both bone and cementum component in a fibrous stroma.

In both cases, treatment done was surgical excision. Both cases are under follow-up, and no recurrence has reported till yet.

DISCUSSION

Cemento-ossifying fibroma is defined by WHO as a demarcated or rarely encapsulated neoplasm consisting of fibrous tissue containing various amounts of mineralized material (bone and/or cementum). Most frequently occurs in female (female: male = 5:1) with age range of 10 to 59 years. They arise in the mandible in 62 to 89% of the patients, 72% occurring in the premolar region. 22% can be found involving molar region of maxilla, ethmoidal and orbital regions and is seen exceptionally in petrous bone. When this arises in children, it has been named the Juvenile aggressive COF, which presents at an earlier age and is more aggressive clinically and more vascular at pathological examination.

Variant of COF differ in nature of calcification (cementum or bone), according to location (oral, paranasal, or orbital), morphological differences (presence or absence of psammomatoid component) and in behavior (aggressive or static). When the lesion contain cementum it is known as cementifying fibroma while presence of bone makes it ossifying fibroma. Lesions with mixture of both cementum and bone matrix are known as cemento-ossifying fibroma. The radiographic appearance is of utmost importance in diagnosis of cemento-ossifying fibroma because it is often needed to separate it from other fibro-osseous lesion. Three radiographic patterns have been described depending on radiographic borders:
1. Defined lesion without sclerotic border (40%).
2. Defined lesion with sclerotic border (45%).
3. Lesion with ill-defined border (15%). In early stages, COF appears as radiolucent lesion with no internal radiopacity. With maturity of the lesion there is increasing calcific flecks progressing ultimately to an extremely radiopaque mass. The growth pattern of the mass is centrifugal so grows equally in all directions presenting therefore as a well-circumscribed mass. Differentiation diagnosis depends on radiographic features of the lesion. It includes—Fibrous dysplasia, cemento-osseous dysplasia, condensing osteitis, Pindborg’s tumors, retained root, odontoma. The boundaries of COF are usually better defined, occasionally have a soft tissue capsule, whereas fibrous dysplasia is usually diffuse and blends with surrounding bone. FD have a characteristic “Ground-Glass appearance” not seen in COF. Also fibrous dysplasia rarely resorbs teeth and the expanded bone still resembles normal morphology. Cemento-osseous dysplasia is usually multifocal, whereas COF is not. Presence of simple bone cyst and a wide sclerotic border is characteristic of cemental dysplasia. Vitality test can help in differentiating COF from condensing osteitis.

Pindborg’s tumors have a high association with impacted teeth. Odontoma can be differentiated by presence of tooth like structures. Retained root will have root canal in root fragment. Multiple COF are rare and there pathogenesis is yet not understood, there are few cases of multiple COF. Familial COF has also been reported.

Yih et al suggested the possibility of a hereditary element. Treatment is surgical in the form of complete resection of the lesion. Since it is low vascularized and well-circumscribed, it is easy to remove it from the surrounding bone. Prognosis is good and recurrence is rare.

CONCLUSION
Diagnosis of cemento-ossifying fibroma is vital because:
1. In initial stages it is completely radiolucent, so it can be confused with other periapical pathologies.
2. As it is usually asymptomatic and has persistent growth potential, it can grow to large size, leading to severe facial asymmetry.
3. And it should not be confused with other fibro-osseous lesion, as their management varies from none to surgical enucleation to complete resection.

REFERENCES