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Cemento-Ossifying Fibroma: Study of Radiographic Features of Six Cases

Cemento-ossifying fibroma is an unusual benign non-odontogenic fibro-osseous tumor that is limited to the jaws and facial bones. Microscopically, some times the lesion may be confused with fibrous dysplasia, and in these cases the final definitive diagnosis requires evaluation of the radiographic configuration.

In this article, the radiological features of six cases of histopathologically confirmed cemento-ossifying fibroma are described.

Keywords: Cemento-Ossifying Fibroma, Fibroosseous Lesion, Radiographic Features

Introduction

Cemento-ossifying fibromas are rare benign, non-odontogenic tumors of the jaw, a subdivision of fibro-osseous lesions. One of its principal characteristics is the massive formation of cementum, cementoid substance or calcified material in the interior of a predominantly fibrous tissue.[1] These tumors occur in the third and fourth decades of life, showing predilection for women.[1-8] The majority of lesions are found in the posterior region of the mandible[1,2,6,8,9] and are usually situated at the roots of the teeth or in the periapical region. Radiographically, the lesion usually has a distinct boundary and in the early stages, it presents as a lucent area. As the lesion matures, bone densities appear, transforming the lesion into a radiopaque mass surrounded by a "halo" of less ossified tissue.[9] They are normally slow-growing and asymptomatic intra-osseous masses[2-4] but a progressive increase in size may eventually cause swelling of the jaw.[9] The growth of the lesion may result in displacement of teeth or the inferior alveolar canal. A significant point is that the outer cortical plate, although displaced and thinned, remains intact. The lamina dura of involved teeth usually is missing, and resorption of teeth may occur.[1] A differential diagnosis should be performed, preferably with other fibro-osseous lesions such as fibrous dysplasia and cemento-osseous dysplasia.[1,10] The term fibro-osseous lesion does not refer to a concrete diagnosis but rather to a group of pathological processes with similar radiological appearances and histopathologies. Some radiographical manifestations may be helpful in differentiating these lesions; however, most published articles present case reports, while surveys of radiographical variations such as internal patterns and effects on the surrounding structures among these tumors have received limited attention. Therefore, in this article we intend to study the radiographic features of six cases of histopathologically confirmed cemento-ossifying fibroma.

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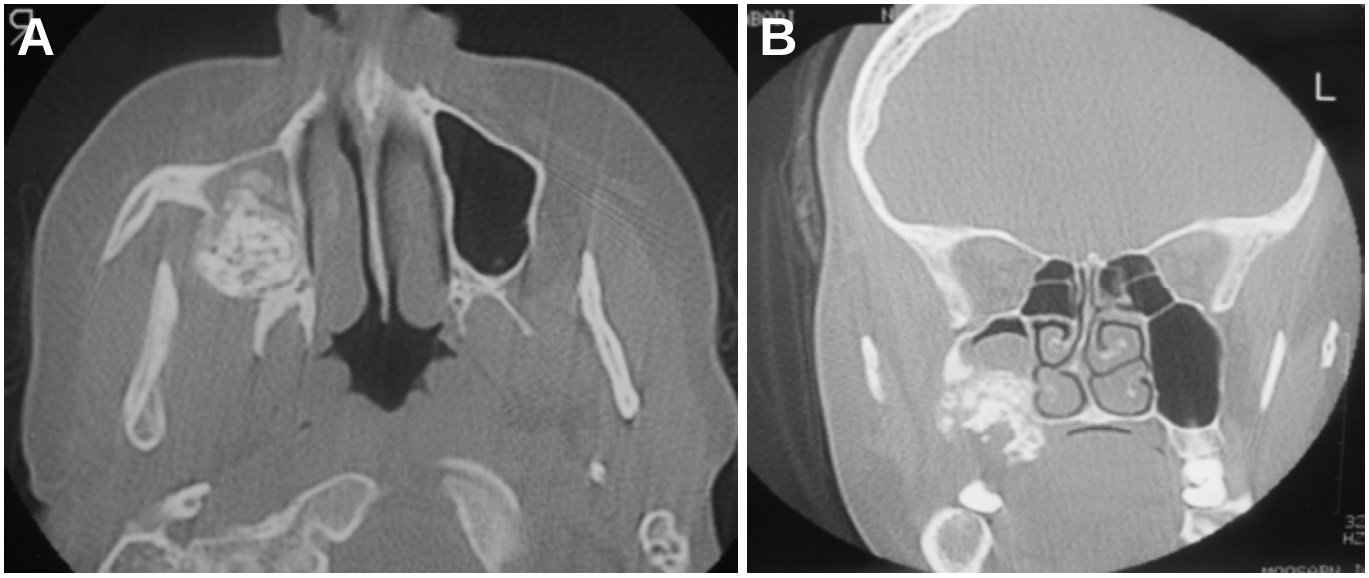

Fig. 1. A 25-year-old woman (case 1) with painful swelling of the right side of the maxilla. The lesion extends into the right maxillary sinus. The peripheral border of the lesion is not parallel to the original shape of the antrum.
A. Axial CT scan
B. Coronal CT scan

Case Presentation

Case 1

A 25-year-old woman was referred to our radiology department complaining of a painful swelling in the right side of the maxilla. Clinically, buccal expansion was evident. In the conventional panoramic radiography, a mixed radiopaque-radiolucent lesion with a thin radiolucent line was revealed. For careful evaluation of the maxillary sinus involvement, axial and coronal sections of computerized tomography of the maxilla were obtained. The border of the lesion was completely well-defined but the outer shape of sinus walls was changed. A cotton-wool appearance in the internal structure was evident (Figs. 1A & B). At the same time, a tissue sample was obtained for a histopathological study that showed benign neoplastic proliferation of fibroblast cells with fibroblastic vascular stroma. In addition, multiple sections of reactive osseous trabeculae and cemental tissues were seen. The anatomopathologic diagnosis was cemento-ossifying fibroma. After one-year of enucleation of the lesion, the patient did not complain of any type of symptoms.

Case 2

An 18-year-old woman complained of a gradually expanding right mandibular swelling from 3 years ago. On clinical examination, a large and painless buccal swelling was evident on the right side of the mandible. The lesion caused no teeth loosening. In the conventional panoramic radiography, a mixed

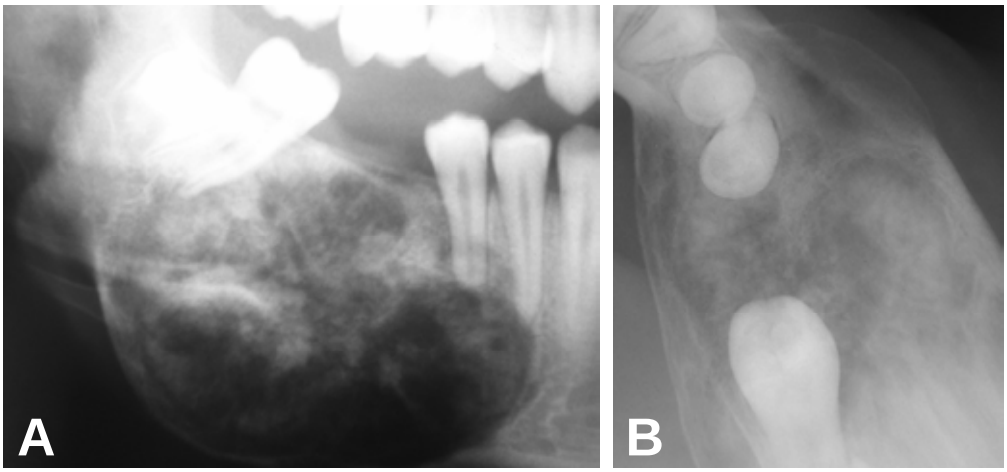

Fig. 2. An 18-year-old woman with expansion in the right mandibular region (case 2).
A. Panoramic view, indicating mixed flocculent appearance of the lesion.
B. Occlusal view; the buccal and lingual expansion is obvious.

Fig.3. A 44-year-old woman with a diastema in the canine and lateral region. The Occlusal view mentions mixed appearance of the lesion.

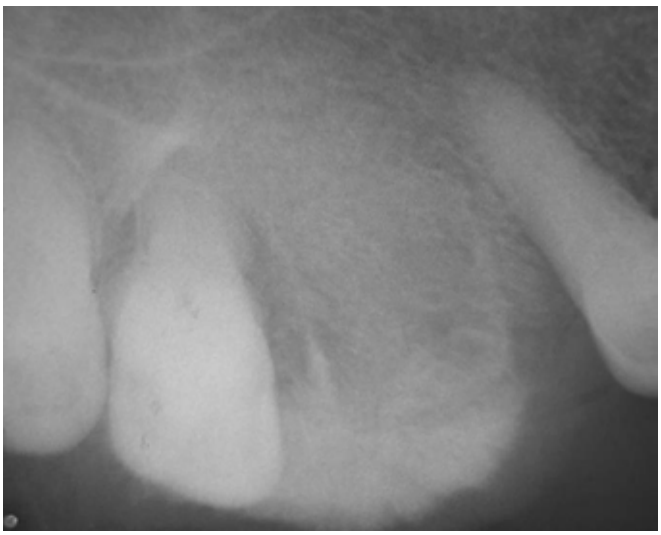

Fig. 4. A 54-year-old woman with history of trauma. Panoramic radiograph indicating the opaque ground-glass appearance related to case 4.

well-defined lesion with an internal flocculent pattern was seen extending from the distal aspect of the right mandibular canine to the mesial aspect of the right first molar tooth causing root resorption of the mandibular right first and second premolar teeth (Fig. 2A). Also displacement of the mandibular second molar and premolar teeth and inferior displacement of the inferior dental canal was obvious. The occlusal view showed buccal and lingual expansion (Fig. 2B). Following an excisional biopsy, the specimen was histopathologically studied. It revealed a fibrous connective tissue with bone trabeculae and spheroidal calcification foci as well as rounded cementicles. The histopathological diagnosis was cemento-ossifying fibroma. After one year of surgery, the patient was asymptomatic and there were no radiographical alterations.

Case 3

A 44-year-old woman presented with swelling and diastema in the left canine and lateral region of the upper jaw. The patient also mentioned a history of a lesion in the same region about 7 years ago, which according to the patient´s file, there was a surgical curettage because of the cemento-ossifying fibroma in that area. Clinically, the hard bony buccal and lingual expansion that extended from the distal aspect of the left second lateral incisor to the mesial aspect of the right first premolar of the maxilla was evident. In the occlusal projection, a radiopaque-radiolucent lesion with a ground glass pattern and a well-defined border was revealed (Fig. 3). Excisional biopsy of the lesion was performed. Histopathological analysis revealed a fibrous stroma with abundant collagen fibers in the sinus in which basophil masses of diverse morphology with areas of dystrophic calcification were found. These findings and the clinical and radiographical study confirmed the diagnosis of cemento-ossifying fibroma. After a one-year evaluation, the patient had no radiographical and clinical alterations.

Case 4

A 54-year-old woman was referred to our department because of swelling in the extraction site of the maxillary right first molar from 2 years ago. The lesion was painless. In palpation, the lesion was bony hard. On the conventional panoramic radiograph, periapical and occlusal projections, a radiopaque lesion in the extraction site of the left first molar was evident. There was a ground-glass appearance in the internal structure (Fig. 4). The enucleated specimen and the incisional biopsy were histopathologically similar. They showed a well-defined, partially encapsulated mass consisting of sheets and bundles of fibroblastic type cells. Occasionally, there were foci of bone with some osteoblastic rimming and rounded foci of cementicles. These features were within the

Fig. 5. A 32-year-old woman with a painless swelling in the mandibular lateral incisor and canine region.
Periapical projection shows the early stages of the lesion with a single radiopaque focus.

Fig.6. A 28-year-old woman with pain and history of trauma. Computerized panoramic radiograph (CR). The ground-glass appearance and bodily displacement of the third molar related to case 6 is evident.

wide spectrum of appearances associated with a cemento-ossifying fibroma. After one year of enucleation of the lesion, the patient was asymptomatic.

Case 5

A 32-year-old woman who complained of a painless swelling in the left mandibular lateral incisor and canine region from 2 months ago was referred to our department. Clinically, a bony hard swelling was evident. In the occlusal projection of the mandible, the cortical expansion and the wispy trabecular pattern was detected. In the periapical view, a radiolucent lesion with a single radiopaque focus and a well-defined border was evident extending from the mandibular left lateral incisor to the left first premolar region (Fig. 5). Following the excisional biopsy, histopathological study was performed; predominantly, a fibrous cellularity and the masses of cementifying tissues with a greater grade of calcification was revealed. After 1-year follow-up of the patient, no recurrent lesion was observed.

Case 6

A 28-year-old woman complaining of simultaneous pain and swelling on the left posterior region of the mandible was referred to our radiology department for evaluation of probable fracture following previous trauma in this region about 4 days ago. A well-defined radiopaque-radiolucent lesion was found incidentally on computerized panoramic radiography (CR). Bodily displacement of the left third molar of the mandible and inferior displacement of the inferior alveolar canal was evident (Fig. 6). On occlusal view, buccal and lingual expansion was seen. Surgical excision of the lesion was performed. Histopathological analysis revealed the presence of fusiform cellular elements arranged in bundles, with variable-size and randomly-distributed amorphous calcifications. The patient was subjected to clinical and radiological follow-up one year after surgical excision of the lesion to discard possible relapses.

Discussion

The age at diagnosis and the sex distribution of patients with cemento-ossifying fibromas (COF) have been reported.[1-12] As in these studies, in our study, COF occurs in the third decades of life (50%), with predilection for women (100%) (Table 1). Although Krausen[13] reported no particular sex predilection and a common occurence in the premolar-molar area of the mandible,[1,2,6,8-12] it has also been reported in the orbital and petromastoid regions, and the maxillary, ethmoidal, frontal and sphenoidal sinuses too.[11,14-21]

In our study, COF occurred in the premolar-molar region of the mandible in 50% of the cases. In some cases, the existence of previous trauma in the area, tooth extractions and prior existence of periodontitis

Table 1. Summary of Significant Clinical Features

Number of Cases	Sex	Age	Location	Pain	History of Trauma
1	F	25	Pos max	Yes	No
2	F	18	Pos man	No	No
3	F	44	Ant max	No	No
4	F	54	Pos max	No	Yes#
5	F	22	Pos man	No	No
6	F	28	Pos man	Yes	Yes

F, Female; Pos max, Posterior maxilla; Pos man, Posterior mandible; Ant max, Anterior maxilla;# extraction of tooth in that area.

have been established as a possible etiological factor.[4,14,19,22,23] In two of our cases, there was a history of previous trauma to the mentioned zone, which is in agreement with their study (Table 1). Clinically, COF presents as a slowly enlarging and asymptomatic lesion,[1-4,22] the diagnosis of which could be confused with Stafne´s idiopathic bone cavity[24] or simulated chronic periapical infectious pathology.[4] In our study, the majority of the lesions had an asymptomatic development, but two patients reported pain and swelling simultaneously. COF may occasionally grow more rapidly and extensively[7] and may even result in mandibular fracture[25]. There are some standout publications regarding the aggressive behavior of these lesions[26-28]. Usually, the bony cortices are expanded and intact in radiographic images[1,9] and perforation of the cortex is rare.[15] The expanding cortex was intact in all the patients. Root resorption and tooth displacement are common findings in these lesions;[1,12] root resorption is seen in 11% and root divergence is recorded in 17% of cases.[11] In the present study, tooth displacement was seen in one case as tilting and as bodily movement in two other cases, but root resorption was seen in one case (Table 2).

Some multiple forms of COF are also described in the literature;[15,29] in this study all of the 6 cases were as a solitary lesion. Some authors have described two basic radiological patterns: a unilocular radiolucency with or without radiopaque foci and a multilocular radiolucent configuration.[11,12] The majority of the lesions in this study were manifested as a well-circumscribed radiolucent-radiopaque lesion and one lesion with a radiopaque appearance (Table 2).

The differential diagnosis of COF includes lesions with a mixed internal structure. Great difficulty may arise in differentiating ossifying fibroma from fibrous dysplasia when the lesion involves the maxillary antrum. Fibrous dysplasia usually displaces the lateral wall of the maxilla into the maxillary antrum, maintaining the outer shape of the wall, whereas an ossifying fibroma has a more convex shape.[1] In this study, in one of the patients, COF extended into the maxillary sinus and the peripheral border of the lesion did not parallel the original shape of the antrum. Also, fibrous dysplasia may change the bone around the teeth without displacing them from an obvious epicenter of a concentrically growing benign tumor.[1] In two cases of the present article (cases 3 and 6), bodily displacement of the teeth from an epicenter together with bone change around them was evident.

Histopathologically, it is typical to encounter a benign fibroblastic stroma with varying cellularity, although mitosis is rare. Within the fibrous stroma there are mineralized tissue masses of basophil aspect that correspond to osteoid or cementoid material.[4] The differences between fibrous dysplasia and COF is not always exhibited in each and every section of the tumor, the reason for which some cases are poorly diagnosed. The histologic pattern of the six lesions coincide in what the authors expressed[4,22] with respect to a predominantly fibrous cellularity and the presence of masses of cementifying tissues, with greater or lesser grade of calcification. MacDonald-

Table 2. Summary of Significant Radiographic Features

Number of Cases	Internal Structure	Root Resorption	Tooth Displacement	Recurrent Lesion	Buccal Expansion	Lingual (palatal) Expansion
1	Mixed cotton-wool	No	No	No	Yes	No
2	Mixed flocculent	Yes	Tilting	No	Yes	Yes
3	Mixed ground-glass	No	Body	Yes*	No	No
4	Opaque ground-glass	No	No	No	No	No
5	Mixed wispy trabecular pattern	No	No	No	Yes	No
6	Mixed ground-glass	No	Body	No	Yes	Yes

*after 7 years

Jankowski[3] has indicated that enucleation or curettage as the first treatment option affords a recurrence rate of 0-28%; if relapse is identified in the course of follow-up, conservative resection is obligate.[30] Although it is a benign neoplasm, recurrences have been reported [7,11,12] and in our study one of the cases was a recurrent lesion occuring after 7 years (according to the patient's file). However, all these patients showed no clinical or radiological evidence of recurrence after one year of post-surgery follow-up.

Based on this study and other studies, it appears that the behavior of ossifying fibroma is unpredictable when radiographic criteria are employed. Some lesions exhibit limited growth potential and may even become arrested after reaching a certain size. Others are capable of accelerated growth potential and recur following surgical curettage. These variations in behavior do not appear to be predictable on the basis of radiographic or microscopic criteria [11,12].

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