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Case Report

Cemento-Ossifying Fibroma of the Mandible in a Young Male Patient- A Rare Case Report with Review of Literature

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ABSTRACT

Cemento-ossifying fibroma (COF) has been recently classified under fibro-osseous lesions of the jaw. It is a relatively rare and benign jaw tumour that has a plethora of histologic appearances, where the cells can differentiate into cementum, lamellar bone, fibrous elements or an admixture of all. The terminology, diagnostic criteria and consensus as to the origin of the tumour is confusing and often controversial; other terms commonly used are ossifying fibroma (OF) and cementifying fibroma. Under the 2017 WHO classification of odontogenic tumours and cysts, it is termed as cemento-ossifying fibroma. This lesion is commonest in the third and fourth decades of life with a female preponderance and commoner in the mandible than in the maxilla. In this article, we describe a rare case of COF presenting as an unusually large, diffuse lytic lesion in the mandible of a 28-year-old male patient.

Key words: Fibro-osseous lesion, cementum, mandible, osteoid

INTRODUCTION

Cemento-ossifying fibroma is a type of rare, benign, fibro-osseous lesion affecting primarily the craniofacial bones especially the jaws, and is of uncertain aetiology. [1] It is a tumour of characterized by proliferation of fibrous tissue, with cementum, bone or a combination of all of these. The striking similarity of this lesion to OF and cemento-osseous dysplasias suggests an odontogenic origin. [2] Menzel was the first to coin the term COF as a variant of OF, in a long-standing tumour of the mandible in a 35-year-old female. [3] The 2005 WHO classification of odontogenic tumours replaced the term COF with OF. [4] The recent update of the WHO classification in 2017 adds the term cement-ossifying fibroma under benign odontogenic tumours of mesenchymal origin. [5] Clinically, these tumours are usually slow-growing and asymptomatic, and do not usually recur if excised. However, in the adolescent age group, they are aggressive and frequently recur; these are termed as 'juvenile ossifying fibroma'. [6] They may be radiolucent but later become radiopaque due to marked calcification, usually seen as a lytic lesion with bone destruction. [7] Histopathologically, they consist of increased amount of fibrous tissue with cementum deposits and osteoid formation. [8] A combination of clinical, radiological

and histological features is highly characteristic for the diagnosis.

CASE REPORT

A 28-year-old male presented with complaint of a slow-growing, progressive swelling over the lower jaw with displacement of teeth. Computed tomography (CT) scan showed a circumscribed lesion with both radiolucent and radio-opaque areas with bone destruction over the anterior and antero-lateral regions of the mandible. A segmental mandibulectomy was done and specimen was sent for histopathological examination.

Gross features: Specimen of segmental mandibulectomy was received measuring 8x6x5cm. A grey-white, firm to hard tumour mass was seen. The cut surface was firm, grey-white and solid (Figure 1). Sections from the tumour were submitted for tissue processing.

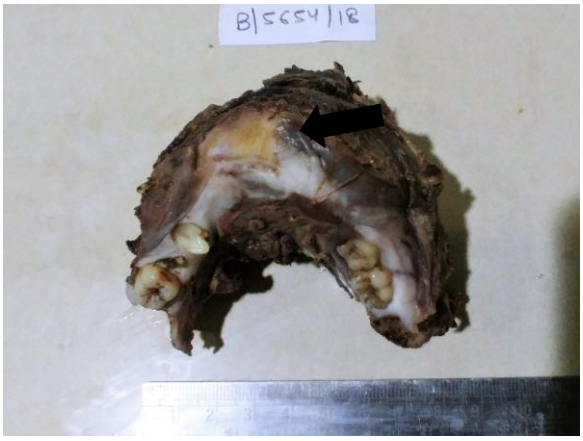

Figure 1: Specimen of segmental mandibulectomy showing grey-white tumour mass over the anterior part of the mandible (black arrow).

Microscopic features: Haematoxylin and eosin stained sections showed tissue lined by squamous epithelium with intense fibroblastic proliferation. Calcified areas showed blobs of cementum-like deposits. Bony trabeculae were also noted along with osteocytes (Figures 2A and B). Chronic inflammatory cells were also seen. A diagnosis of cemento-ossifying fibroma was made.

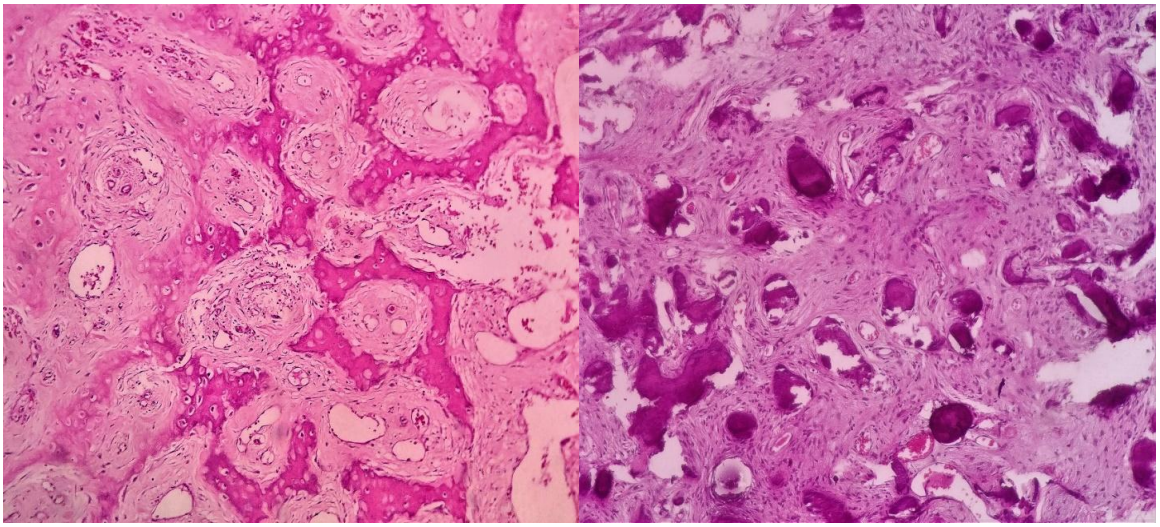

Figure 2A: Section shows fibroblastic proliferation along with trabeculae. Figure 2B: Shows cementum-like deposits often in a concentric fashion resembling psammoma bodies.

DISCUSSION

COF is a benign fibro-osseous lesion. [9] Though of uncertain origin, they are thought to arise from the periodontal ligament as they are able to produce cementum and osteoid. [10] Trauma has also been suggested as a cause for COF [11-12] but there was no such history in our case. WHO 2017 classifies it as a benign odontogenic tumour of mesenchymal origin. This terminology is also helpful to distinguish it from the juvenile variant. This tumour is five times more common in females. About 62 to 89% of cases occur in the mandible.

[13] It is rarer and more aggressive in the maxilla. [14] Tumours larger than 80 mm in size have been term as giant ossifying fibromas. Radiologically these tumours have distinctive features; they can be unilocular or multilocular and are initially radiolucent. With time, there is increasing calcification and it is visualized as a radio-opaque mass. Importantly, the tumour grows in all directions and usually does not cause any root resorption of tooth. [6] Another important feature is that COF is usually well-circumscribed. [10] In the present case, sclerotic margins with both radio-opaque and radiolucent areas were seen, suggesting progressive calcification and long duration of the tumour. There was absence of significant clinical symptoms other than long-standing painless swelling and mild facial deformity.

Histologically, the tumour shows highly vascular and cellular fibrous tissue stroma with areas of concentric cementum-like deposits, along with formation of osteoid and anastomosing bony trabeculae surrounded by fibroblasts. [15]

Differential diagnoses of COF include fibrous dysplasia, odontoma, osteosarcoma, ossifying fibroma, Pindborg tumours, odontogenic keratocyst and adenomatoid odontogenic tumour. [16] Fibrous dysplasia, the closest differential, has ill-circumscribed margins unlike COF. Absence of rapid onset of symptoms eliminates the presence of inflammatory lesions. Osteosarcoma can be differentiated from COF by distinct radiological signs and marked atypia on microscopy. Odontomas show the presence of dentin, which is similar to bone but show tubule-like structures, and enamel which has a 'fish-scale' appearance. [5]

CONCLUSION

The term cement-ossifying fibroma is thus given to a distinct form of fibro-osseous lesion which contains both types of tissue and because of the difficulty in reliable histopathological differentiation between immature bone and cementum. It is often discovered incidentally. COF is usually a sharply circumscribed lesion and complete excision is the preferred treatment. [6] There are reports of recurrence, [1-2,10] but this has not been observed in our case. COF should be considered as a possible differential in slow-growing lesion of the jaw in females, though our case was a male patient. Multidisciplinary approach with correlation between clinical, radiological and histopathological diagnoses is crucial for the diagnosis.

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