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Cemento-ossifying fibroma involving mandible – A rare case report

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Abstract

The Cemento-ossifying fibroma (COF) is a benign mesenchymal odontogenic neoplasm of the bone characterized by the replacement of normal bone by fibrous tissue and varying amounts of osteoid and cementum-like material. The definitive diagnosis of such lesions requires clinical, radiological, and histo pathological observations. The treatment of choice is surgical resection or enucleation according to the size of the lesion. Recurrence after complete removal of the lesion is rare. We hereby report a case of COF in the left mandibular premolar region with detailed clinical, radiological and histopathological features along with its differential diagnosis.

Keywords: fibro-osseous lesions, cemento-ossifying fibroma, Ossifying fibroma, Brush-border

Introduction

Fibro-osseous lesions are a heterogeneous group of benign lesions of unknown etiology affecting the jaws and other craniofacial bones, which are characterized by the replacement of normal bone by fibrous tissue, and contain newly formed, mineralized product. [9]

The concept of 'fibro-osseous lesions' of bone has evolved over the last several decades and now includes two major entities: fibrous dysplasia and ossifying fibroma of jaw as well as the other less common lesions such as florid osseous dysplasia, periapical dysplasia and focal sclerosing

Osteomyelitis. [1] The cemento-ossifying fibromas have been described as well demarcated or rarely encapsulated neoplasms, consisting of fibrous tissue containing varying amounts of mineralized material resembling bone and/or cementum [1,4]. However, various terms like ossifying fibroma, cementifying fibroma, cemento-ossifying fibroma and central fibro-osteoma are continued to be used by various authors.

It mostly affects patients between the third and fourth decades of life, with a prominent female predilection. (Female: male, 4:1). Among the craniofacial bones, mandible is mostly affected at the molar premolar area [2,5]. Clinically, these Tumors manifest as a slow enlarging intra-bony mass that is normally well delineated and asymptomatic. Though it may become large enough over time to cause pronounced facial asymmetry.[2]

Menzel gave the first description of a variant of ossifying fibroma, and named it as 'cemento-ossifying fibroma' in the year 1872. Almost after a centennial in 1992, the World Health Organization (WHO) revised the nomenclature, and the separate lesions of 'cementifying fibroma' and 'ossifying fibroma' were clubbed together to form a single entity of "cement-ossifying fibroma". Again, the term "cementoossifying fibroma" was issued by the fourth edition of the WHO classification of 2017, describing it as a benign mesenchymal odontogenic tumour. [5]

The radiological aspect of 'cemento-ossifying fibroma' is initially characterized by a well -defined lesion that is easily distinguished from the surrounding bone. At an early stage, the lesion is well circumscribed, radiolucent in nature. As it evolves, thin opacities develop at the canter of the lesion; which have lower density than that of the surrounding bone. At a more advanced stage of maturation, it shows irregular opacities formed by mineralized tissue. With time due to increased calcification the lesion often observed as nearly complete radioopaque mass. [10]

Histopathologic ally, these Tumors are composed of well vascularized fibro- cellular tissue with the capacity to form varying types of mineralized product which include an admixture of osteoid, bone, and basophilic acellular cementum-like spherules. The bony trabeculae vary in size and frequently demonstrate both woven and lamellar patterns. Peripheral osteoid and osteoblastic rimming are usually present. The cementum-like spherules often demonstrate brush borders that blend into the adjacent connective tissue. [2,9]

Surgical removal and curettage is the treatment of choice. However, surgery of large lesions may result in massive tissue ablation and it is difficult to rehabilitee the affected region. The prognosis is usually good. Recurrences of ossifying fibromas are variable, with a range of 10 -25% after enucleation and 5 % after resection.

We hereby report a case of COF in the left mandibular molar-premolar region with detailed clinical, radiological and histopathological features.

Case report

A 39-year-old female patient reported to the Department of Oral & Maxillofacial Pathology, Guru Nanak Institute of Dental Sciences & Research (GNIDSR), Kolkata with a complaint of a swelling in the 36 region of left mandible for past 7 years.

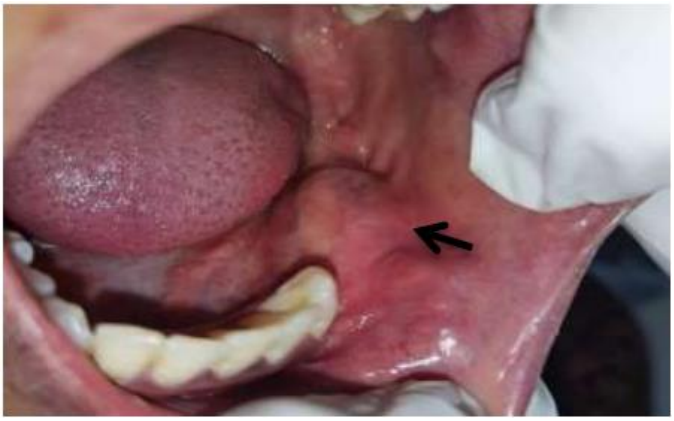

Fig 1: Presence of diffuse swelling in the left mandibular region. (Black arrow)

Intraoral examination revealed a diffuse, bony hard, nontender swelling of the left mandibular buccal cortex extending from mesial aspect of 35 to distal aspect of 37, causing obliteration of the buccal vestibule. [Fig 1]

Orthopantomogram (OPG) revealed a well-demarcated unilocular radiolucent lesion interspersed with areas of radiopacity extending anteriorly from mesial aspect of missing 35 up to distal aspect of 37 posteriorly. [Fig2] Cone Beam Computer Tomography revealed well defined lesion of mixed radio density involving body of the mandible at the molar premolar area. [Fig3]

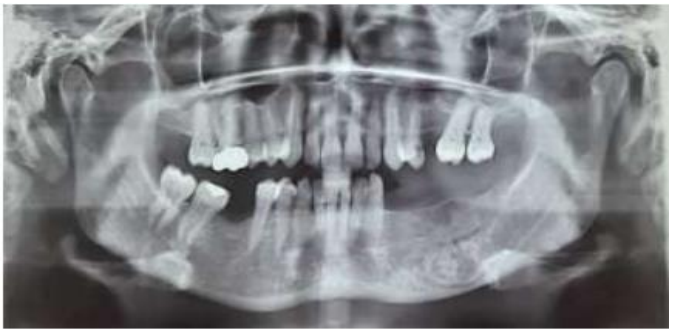

Fig 2: OPG reveals mixed degrees of radio-opacities on the left mandibular premolar region

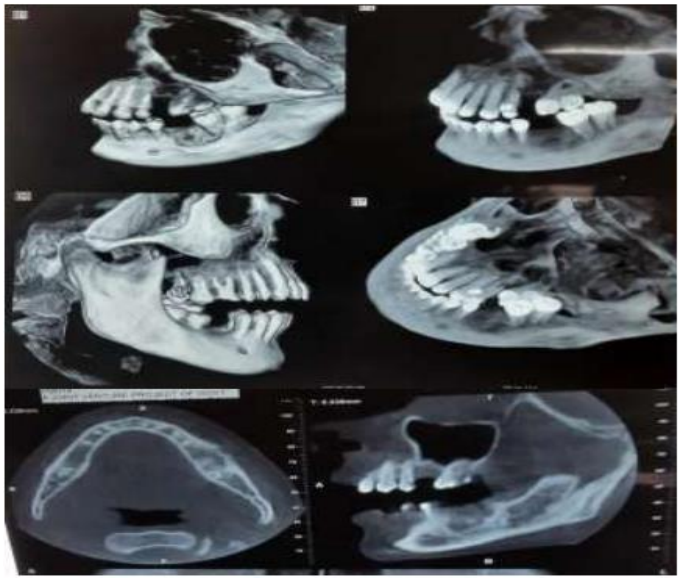

Fig 3: CBCT revealed well defined lesion of mixed radio density involving body of the mandible

An incisional biopsy was performed from the representative site of the lesion under local anesthesia and the tissue was subjected to histopathological diagnosis.

Histopathological examination revealed the presence of cellular fibrous connective tissue with multiple diffuse irregular basophilic areas with varied degrees of calcification. Few of the calcified areas revealed Irregular brush border at the periphery blending into the connective tissue. [Fig 5]

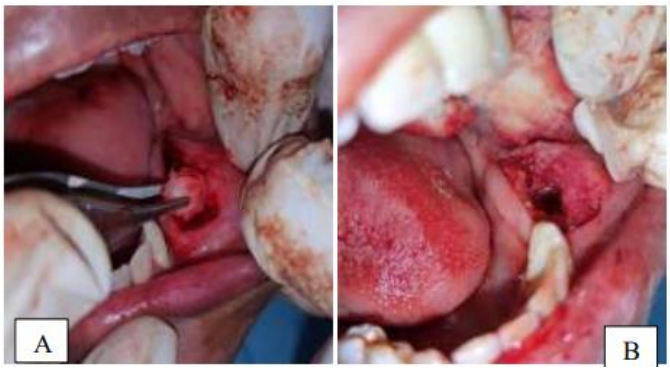

Fig 4: A&B paroperative picture revealed d retrieval of biopsy specimen.

There is also presence of numerous basophilic, acellular spherules resembling cementum and multiple

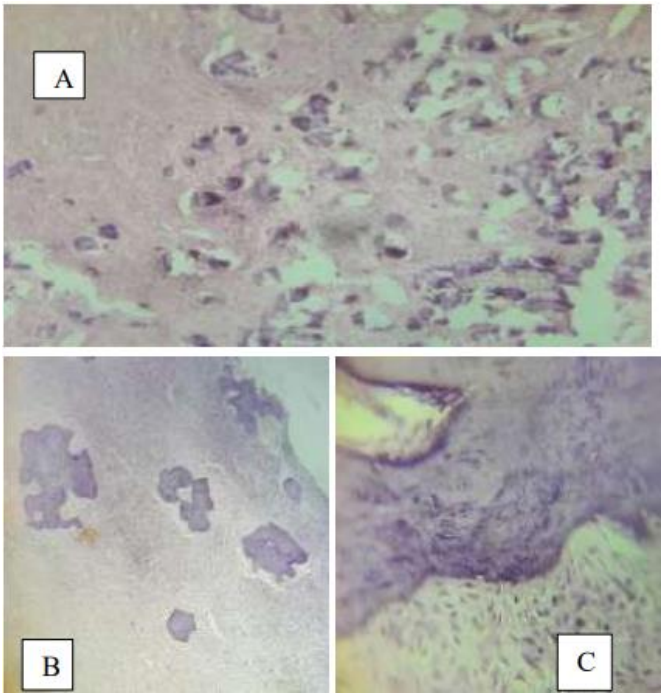

Fig 5: (A, B, C) H & E photomicrograph revealed presence of fibro cellular connective tissue stroma with varying degrees of calcified materials. With presence of numerous basophilic acellular spherules resembling cementum like material (4x,10x ,40x)

basophilic irregular trabecular masses of varying size throughout the connective tissue with few osteocytes entrapped within them resembling woven / immature bone. The basophilic areas is supported by interlacing collagen fibers in a background of intense proliferating plump mesenchymal cells resembling active fibroblasts. The overall histopathological features are suggestive of "Cemento-Ossifying Fibroma.

The patient was further referred to the Department of Oral & Maxillofacial surgery for further management.

Discussion

Cemento-Ossifying Fibroma (COF) is a relatively rare, benign, non-odontogenic neoplasm affecting the bones of the craniofacial structure. [5]

Prakash et al reported that though it can occur in any part of the facial skeleton, 70-90% of the head and neck lesions is seen in the mandible. The present case also revealed the same site of occurrence.[7] Clinically these lesions present as a slow growing painless mass and inconspicuous in nature until the swelling of the face becomes clearly evident. According to various literatures in few cases, the lesion can grow aggressive growth pattern. However in our case the lesion was slow growing which has attained the present size (4cm x3 cm) over the past seven years. Recently the patient encountered occasional bouts of mild pain at the site of occurrence. [7]

Radiologically COF exhibit well defined mixed radiolucent and radiopaque mass Bandura et al, reported that the lesion radiologically can present as a well-defined multinodular mixed radiolucent and radio-opaque mass with marginal sclerosis, which differentiates it from fibrous dysplasia. Similar radiologic features were also noted in the case discussed here. [6]

Provisional diagnosis of cemento-ossifying was made on the basis of clinical and radiographic features. The differential diagnosis included fibrous dysplasia, cemento-osseous dysplasia, calcifying epithelial odontogenic tumor, (CEOT) florid cemento-osseous dysplasia, and odontometer. An ossifying fibroma is a benign tumour in bone having a distinct margin whereas fibrous dysplasia is a maturation defect of bone which blends into the adjacent normal bone. which was not seen in the present case.

On the other hand, cemento-osseous dysplasias show the presence of wide sclerotic border, which it is not corroborative to COF. Odontoma is characterized by presence of clusters of tooth-like structure in the lesion, whereas COF shows radiopaque foci of calcified areas. Radiographically CEOT presents with driven snow appearance which sometimes may mimic COF making it difficult to diagnose. Thus histopathological evaluation

is necessary to differentiate the aforementioned lesions. [6,10]

Aburas et al stated that this bone tumor consists of highly cellular, fibrous tissue having varying amounts of calcified structure resembling either bone, cementum or both. Similar features were also noted in our case. In case of CEOT sheets of polyhedral neoplastic odontogenic epithelial cell along with well-developed intercellular bridges are seen which was not evident in our case.

Depending upon the clinical, radiological and histo pathological feature the case was diagnosed as 'Cemento – Ossifying Fibroma.

Due to confined nature of the tumor Javier et al stated in their study that surgical removal and curettage is the treatment of choice. [1]

However, after proper diagnosis the patient was referred to department of Oral and Maxillofacial Surgery for necessary action. The recurrence of these benign Tumors following surgery is very rare, though Eversole and his co-workers in a study of 64cases of cemento-ossifying fibroma reported a recurrence rate of as high as 28 per cent following surgical curettage.

Conclusion

As there is no distinctive features for COF it needs proper correlation of clinical, radiological and histopathological features to reach an accurate diagnosis. Curettage or enucleation is indicated for smaller lesions, while larger lesions need surgical resection with a safety margin. Prognosis is favourable without any obvious potential for malignant transformation

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