

Demystifying the Mystified - An Ambiguous Presentation of Cemento-ossifying Fibroma in Maxilla

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Authors' contributions

This work was carried out in collaboration between all authors. Authors NR, DAT and RSR studied the case and evaluated. Authors NR and DAT designed the case study and wrote the first draft of the manuscript. All authors managed the literature searches. Author MR managed histopathology report. All authors read and approved the final manuscript

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Case Study

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ABSTRACT

Cemento-ossifying fibroma is a slow growing benign tumor of mesenchymal origin. It is typically found in craniofacial bones, frequently involving the mandible and it tends to occur in third and fourth decade of life with predilection for women. Occurrence of this lesion in maxilla is a rare entity. We report one such case of cemento-ossifying fibroma of size 5x4 cm on upper left maxilla in 38 year old female patient which is an unusual presentation.

Keywords: Fibro-osseous lesion; Cemento-ossifying fibroma (COF); maxilla; benign tumor; mixed radiolucent -radiopaque lesion.

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1. INTRODUCTION

Fibro-osseous lesions are a diverse group of diseases that are characterized by replacement of normal bone by fibrous tissue containing variable amount of mineralized material [1]. Ossifying fibroma is categorised under benign fibro-osseous lesion along with fibrous dysplasia, osseous dysplasia. Despite the similarities in clinical, radiographic and histopathological features, these lesions are of distinctive nature [2].

Ossifying fibroma is a benign neoplasm usually present as a painless, slow growing expansile lesion affecting the jaws [3]. Cemento ossifying fibroma is a fibro-osseous lesion that arises from periodontal ligament whereas ossifying fibroma is of bony origin [4].

Most of the time these lesions are slow growing, asymptomatic and usually unnoticed by the patient until the swelling of the face is noted. Nevertheless, few lesions may grow rapidly and cause symptoms. This part of the lesion hinders the quality of life of the patient. Therefore a proper diagnosis and treatment plan is required to achieve a good result. The present case depicts the rare entity of occurrence in the maxilla in an adult female patient.

2. CASE PRESENTATION

A 38 years old female patient presented to the Department of Oral medicine, Oral Diagnosis and Oral Radiology, Vishnu Dental College, Bhimavaram, with the complaint of swelling over the left cheek region. As per the patient, the swelling had started 9 months earlier from the day of initial visit. Patient reports that there was a gradual increase in size of the swelling with no history of pain. There was no past history of trauma associated with the swelling as reported by the patient.

This condition does not seem to have affected other family members and history of no known allergies as reported by the patient.

General physical examination showed the patient to be conscious and well oriented. Clinical examination revealed gross facial asymmetry due to swelling on the left side of the face measuring about 5x4 cm seen extending superiorly 0.5 cm below the infraorbital ridge and inferiorly 2 cm above the inferior border of the mandible and mediolaterally extending from the

corner of the mouth to 2 cm from the angle of the mandible (Fig. 1) Skin over the swelling showed normal colour with stretched contour. On palpation the swelling was non tender and hard in consistency.

Intraoral examination revealed a solitary swelling involving left maxilla measuring 5x3 cm, extending anteriorly from first premolar region to the maxillary tuberosity posteriorly, medially 0.5 cm from the mid palatine raphe and laterally causing obliteration of the buccal vestibule (Fig. 2). The surface of the swelling was smooth with no signs of discharge. On palpation the swelling was non tender and hard in consistency. Other findings over the adjacent area of the lesion includes displacement of second and third molar.



Fig. 1. Extra oral picture showing swelling over left cheek (Both frontal and lateral view)



Fig. 2. Intra oral picture showing lesion involving left maxilla

Based on the history and in correlation with the aforementioned clinical findings, a provisional diagnosis of a benign fibro-osseous lesion involving left maxilla was made, and the differential diagnosis of fibrous dysplasia, odontogenic myxoma, cemento-ossifying fibroma was considered.

Patient was further subjected to laboratory and radiological investigations. This included serum calcium, phosphorus and alkaline phosphatase levels along with routine haematological investigations. All the laboratory investigation parameters reported to be within the normal limits.

Radiological investigations include intra oral periapical radiograph, orthopantomograph and computed tomography (CT). Intra oral periapical radiograph showing 24, 25, 26 tooth region with internal structure being mixed radiolucent-radiopaque lesion (Fig. 3). Orthopantomograph showed a well defined radiolucent lesion with flecks of radiopaque foci seen involving the left maxillary posterior region extending from first premolar to the maxillary tuberosity. Superiorly involving the floor of maxillary sinus and inferiorly involving the alveolar ridge with displacement of second and third molar (Fig. 4). For better localization of the lesion advanced imaging CT is performed. Coronal section revealed a well circumscribed expansile mass lesion involving left maxilla of size approximately 5.5 cm x3.4 cm and internal structure depicted mixed radiographic image with variable amount of predominant radiopaque foci and radiolucent areas. 3D CT imaging was performed which represents tumor extension with encroachment into surrounding vital structures (Figs. 5a, 5b).

An incisional biopsy was performed from the site of the lesion. The histopathological report of the specimen revealed hypercellular connective tissue with areas of ossification and areas of woven bone with osteoblastic rimming and osteocytes in the lacunae at the centre (Figs. 6a, 6b).



Fig. 3. Intra oral periapical radiograph depicting the lesion in the premolar and molar region



Fig. 4. Orthopantomograph depicting the lesion in left maxilla

Owing to the histopathological features and correlated with clinical and radiographic findings a final diagnosis of cemento-ossifying fibroma of left maxilla was made.



Fig. 5: a: Coronal section, b: Three dimension CT scan revealing the lesion extensions

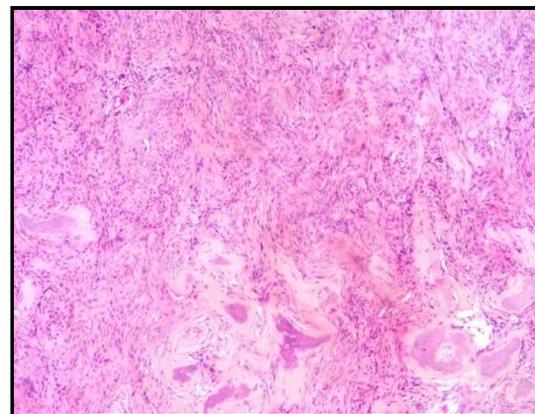


Fig. 6. 10 x view of H&E stained section showing areas of osteoid formation within an active fibrocellular stroma

3. DISCUSSION

Cemento-ossifying fibroma (COF) is a fibro-osseous lesion which contains multipotent cells

that are potential of forming cementum, lamellar bone and fibrous tissue [4,5]. It has been documented that pluripotent mesenchymal cells could differentiate to produce calcified material resembling cementum and bone [6]. Despite its origin is from periodontal membrane, the factors that stimulate this structure to produce cementum in an aberrant anatomical site remains controversial. Inflammation secondary to infections or trauma has been proposed as a causative agent [7].

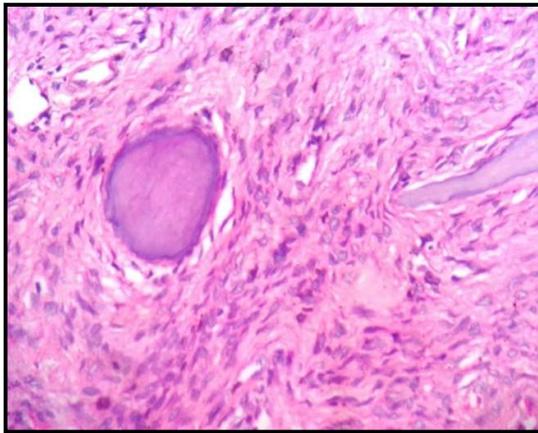


Fig. 7. 40 x view of H&E stained section showing area of basophilic spherule of cementum

Clinically cemento-ossifying fibroma usually presents as a painless slow growing expansile lesion where swelling with displacement of the teeth may be the first clinical feature as seen in the current case. This lesion is more common in females in the third and fourth decade of life [3]. Many studies reported that commonest site of occurrence is posterior region of the mandible. Very few cases have been reported in maxilla as in the present case which remains as a rare

entity. Regarding the age of onset of cemento-ossifying fibroma, the present lesion is in accordance with the majority of the literatures (Table 1).

The radiographic appearance is of prime importance in the diagnosis of cemento-ossifying fibroma because it is often needed to differentiate it from other fibro-osseous lesions. In the early stages, cemento-ossifying fibroma appears as a radiolucent lesion with no evidence of internal radiopacities. As the tumor matures, there is increasing calcification so that radiolucent area becomes flecked with opacities until ultimately the lesion appears as an extremely radiopaque mass. Majority of them present as well defined mixed density lesions, with very few being completely radiolucent. Resorption of roots and divergence of associated teeth with loss of lamina dura may be noted [13]. In the current case, the lesion showed mixed radiodensity causing tooth displacement and loss of lamina dura.

There are three different patterns of radiographic borders which are defined lesion without sclerotic border (40%), defined lesion with sclerotic border (45%), ill-defined borders (15%) indicating rapidly growing tumor [6]. In the current case the lesion is well defined with sclerotic border.

Radiographically fibrous dysplasia is the main lesion considered in the differential diagnosis of cemento-ossifying fibroma (COF) [14]. Fibrous dysplasia usually appears as diffuse homogenous ground glass radiodense lesion blending with the surrounding bone. One additional important radiographic diagnostic feature of cemento-ossifying fibroma is that there is a centrifugal growth pattern rather than a linear

Table 1. Clinical and radiographic features of few reported cases in the literature [8,9,10,11,12]

Authors & years	Age & Gender	Location	Radiographic features
Tamiolakis D, et al.[8] (2005)	36 yr/female	Maxilla	Mixed radiolucent & radiopaque
Liu Y, et al. [9] (2010)	20 yrs/female	Maxilla	Completely radiopaque lesion
Sarwar HG, et al. [10] (2010)	11 yr/male	Maxilla	Mixed radiolucent & radiopaque
Siddana Gouda S, et al. [11] (2014)	22 yr/female	Maxilla	Completely radiopaque lesion
Mithra R, et al. [12] (2015)	32 yr/female	Maxilla	Mixed radiolucent & radiopaque
Present case	38 yr/female	Maxilla	Mixed radiolucent & radiopaque

one and therefore these lesions grow by expansion equally in all directions which produces a round tumor mass [10]. This characteristic presentation is evident in the present case. Among osseous dysplasias early, intermediate and late stages of focal osseous dysplasia (FOD) could be considered in the differential diagnosis [3]. FOD is mostly seen in patients during fourth and fifth decades of life and smaller in size than ossifying fibroma and usually presents with ill defined borders.

Histopathology reveals many delicate interlacing collagen fibers, arranged in discrete bundles, interspersed with large numbers of active, proliferating fibroblasts and cementoblasts. As the lesion matures, the islands of calcification increase in number, enlarge, and ultimately coalesce. The increase in degree of calcification, accounts for the increase in the radiopaqueness of the lesions in the radiograph [10]. Microscopic features of the case presented above showed characteristic calcified material with in fibrous connective tissue stroma which is an indicative feature of cemento-ossifying fibroma.

The treatment consists in completely removing the lesion with curettage, surgical excision or en-block resection, depending on the size, location and extent of the lesion. COF is usually well circumscribed and this facilitates its extirpation from the surrounding bone and recurrence is rare and death resulting from this tumor is very rarely reported [8]. In lesions with a fibrous capsule around, the surgical excision may be performed more easily [3,10]. In the present case complete excision of the lesion was planned.

4. CONCLUSION

Cemento-ossifying fibroma of the maxilla is an uncommon benign tumor with more female predilection. Since the lesions can be diagnosed based on clinical and radiological findings to a major extent, the role of oral physician and oral radiologist always remains challenging in arriving at a proper diagnosis and providing a proper and definite treatment plan thus ultimately improving the quality of the life of the individual. A correlation between clinical, radiographic and histological features is the key to establish the correct diagnosis.

CONSENT

All authors declare that 'written informed consent was obtained from the patient for publication of this case report and accompanying images.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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