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Dr. Mahendra Patait
Professor in Head, Department of OMDR, SMBT Dental College, Sangamner, Maharashtra, India

Dr Vishakha Chaudhari
PG Student, Department of OMDR, SMBT Dental College, Sangamner, Maharashtra, India

Dr. Ashok Mathew
Assistant Professor, Department of Surgical Sciences, University of Science and Technology, Fujairah

Dr. Rajan Mangrolia
Senior Lecturer, Department of Conservative Dentistry and Endodontics, SMBT Dental College, Sangamner, Maharashtra, India

Dr. Mahesh Ahire
Head of Department, Department of Periodontology, SMBT Dental College, Sangamner, Maharashtra, India

Corresponding Author:
Dr. Mahendra Patait
Professor in Head, Department of OMDR, SMBT Dental College, Sangamner, Maharashtra, India

Cemento-ossifying fibroma: Case report

Dr. Mahendra Patait, Dr Vishakha Chaudhari, Dr. Ashok Mathew, Dr. Rajan Mangrolia and Dr. Mahesh Ahire

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Abstract
Ossifying fibroma (OF) is a benign, non-odontogenic tumor of the jaw, and it is classified as fibro-osseous lesion of jaw. Earlier, the lesion was sub classified histologically into ossifying fibroma and cementifying fibroma according to the hard tissues formed either bone or cementum. Menzel in 1872 called it as cemento-ossifying fibroma (COF). This lesion is included in the group of mesodermal odontogenic tumors and usually present as a gradually growing lesion that might achieve enormous size with resultant deformity, if not treated. The tumor may grow quite extensively; thus, the term "aggressive" is occasionally applied. In this article, A case of cemento-ossifying fibroma involving the left side of mandible is described in a 25 year-old female patient. The clinical, radiographic, histologic features are described and several differential diagnoses are discussed.

Keywords: Ossifying fibroma, cemento-ossifying fibroma, aggressive tumor, diagnosis

Introduction
Menzel, gave the first opinion of a variant of ossifying fibroma, calling it a cement-ossifying fibroma in 1872. Cemento-ossifying fibroma (COF) is a distinct form of benign fibro-osseous lesions of the jaw, affecting primarily the craniofacial region. The World Health Organization (WHO) initially classified Cemento-ossifying fibroma as a fibro-osseous neoplasm. Nevertheless, they do not arise in the long bones, and occur mostly in the tooth-bearing areas of the jaws [1, 2] WHO in 1972, considered ossifying fibroma to be a tumor of the bone origin and cementifying fibroma as a tumor of the odontogenic origin [3] However, in 1992, WHO grouped such lesions under the common denomination of Cemento-ossifying fibroma on the grounds that they correspond to histological variants of a same type of lesion. In the past, many investigators discretely classified cementifying fibromas and ossifying fibromas. When wavy trabeculae or spheroidal calcifications were encountered, the lesion was often referred to as cementifying fibroma. When the bone predominated, ossifying fibroma was assigned. Today, however, the term 'cemento-ossifying fibroma' is widely used because both osseous and cemental tissues are seen commonly in a single lesion. According to WHO, Cemento-ossifying fibroma is classified as fibro-osseous lesion or non-odontogenic tumor of the jaws [4]. The lesion has some similarity to ossifying fibroma and cemento-osseous dysplasias; This gives substantiation of their odontogenic origin. Some authors stated that they are derived from the mesenchymal cells of the periodontal ligament; these cells have a tendency to form fibrous tissue, cement and bone or a combination of such elements. It was reported by Ever sole *et al*. that these cementum-like structures are associated with the membranous bone, and may not be related to cement genesis. While it is also believed that they developed from the remnants of cementum in the tooth socket after extraction of the tooth. Cemento-ossifying fibroma has always been bounded by controversy regarding the expressions and criteria of diagnosis.

Case Report
A 25-year-old female reported to the outpatient department of Oral Medicine, Diagnosis and Radiology with a slowly progressive painless swelling over left side of jaw since 2 years.

The extraoral examination does not show any noticeable swelling, the regional lymph nodes were nonpalpable and non-tender, with no signs of inflammation. The overlying skin was intact and no paraesthesia was noted on the surrounding structures [Figure 1]. Intraoral examination revealed a localized swelling was seen on left side of jaw, extending from the distal aspect of canine to the distal aspect of first premolar region with buccal cortical plate expansion obliterating the buccal vestibule. The involved teeth showed displacement. The overlying mucosa appeared to be normal. On palpation the swelling was nontender and bony hard in consistency [Figure 2]. The involved teeth were vital. All the vital signs were within the normal limits. Based on clinical findings, a provisional diagnosis of Fibro-osseous lesion of left mandible was prepared. Intraoral periapical radiograph of mandibular left premolar region, and orthopantomograph [Figure 3 and 4] revealed a single, large, well-defined mixed radiolucent –radiopaque lesion resembling cyst like appearance, involving the left body of mandible, extending interdentally from the space between the canine and first premolar region anterioposteriorly and superioinferiorly involved the alveolar crest to approximately 3-5 mm below the tooth apices with the displacement of involved tooth without root resorption. The coronal, sagital and an axial 3D reconstructed computed tomographic images [Figure 5, 6 and 7] revealed a well-defined, heterogenous, expansile, dense calcifying lesion of lower left mandibular body, extending into the interdental space between left canine and first premolar teeth region of size 17.6×23.3 mm with cortical plate thinning. Routine hemogram and serum chemistry (serum alkaline phosphatase and serum acid phosphatase) values were within normal range. Incisional biopsy was performed. It was reported a well-circumscribed and partly encapsulated lesion consisting of an abundant cellular fibrous tissue with scattered trabeculae of lamellar bone, metaplastic bone and cementoid masses. The stromal cells were spindle shaped and had bland-looking nuclei. These findings were consistent with Cemento-ossifying fibroma. The lesion was surgically excised as per the traditional approach [Figure 8 and 9] and the excised tissue was sent for the histopathologic examination which was suggestive of Cemento-ossifying fibroma. The patient was kept under follow-up

Fig 1: Extraoral presentation

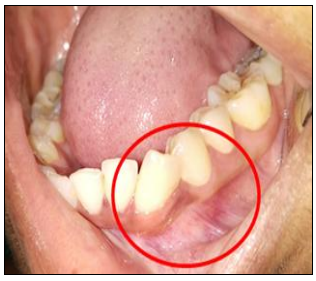

Fig 2: Intraoral presentation

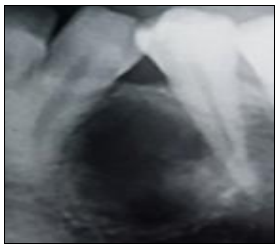

Fig 3: Intraoral periapical radiograph of mandibular left premolar region, showing well defined unilocular mixed radiopaque radiolucent lesion.

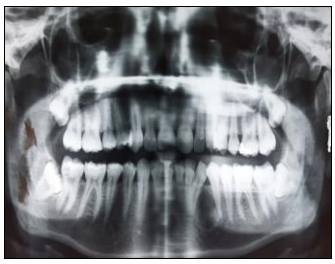

Fig 4: The orthopantomograph the white arrow discovered a single, unilocular, well defined mixed radiolucent – radiopaque lesion resembling cyst like appearance.

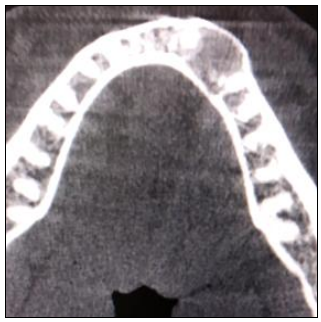

Fig 5: The cone beam CT, the axial section revealed a well-defined, corticated, expansile lesion with one or two rounded opacities inside the radiolucency of lower left mandibular body

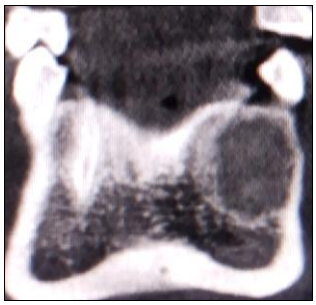

Fig 6: The cone beam CT, The coronal section showed a well define single unilocular radiolucency surrounded by smooth corticated radiopaque border on left side of body of mandible.

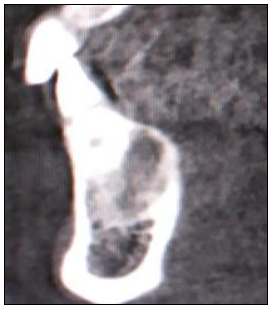

Fig 7: The cone beam CT, The sagital section exposed the unilocular mixed radiopaque radiolucent lesion having corticated borders at the level of left mandibular canine

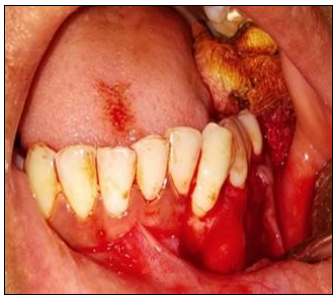

Fig 8: The intraoperative view, the complete excision of lesion from buccal aspect between the left mandibular canine and first premolar teeth region.

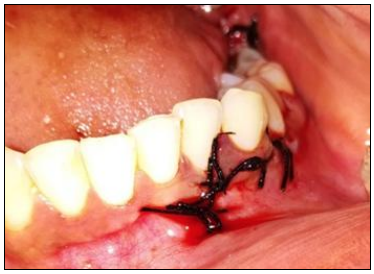

Fig 9: The postoperative view, the silk sutures were placed after complete excision of the lesion.

Discussion

The cemento-ossifying fibroma has been mentioned to as osteo-fibroma, fibro-osteoma and benign fibro-osseous lesion originated from the mesenchymal cells of periodontal ligament. Pindborg, Waldron and many other authors has been discussed the concept, that the central ossifying fibroma and Central cementifying fibroma shows similar clinical, radiographic and histopathological features. For this reason depending on their origin it has been recommended that these two are discrete entities with cells undergoing proliferation; osteoblast with bone formation in central ossifying fibroma and cementoblast with cementum formation in central cementifying fibroma. Today, the term 'cemento-ossifying fibroma' is generally used because both osseous and cemental tissues are seen ordinarily in a single entity. The Cemento-ossifying fibroma is classified as fibro-osseous lesion or non-odontogenic tumor of the jaws according to WHO [3, 4]. The current concepts of origin of cemento-ossifying fibroma include traumatic and developmental causes. Cakir and Karadayi reported nasopharyngeal origin from the embryologic nests [4] Brademann *et al*. explained that ectopic periodontal membrane differentiating from primitive mesenchymal cells serve as a cause of development of COF in the petrous bone, and trauma may be a principal factor in the stimulation of proliferation of cemento-ossifying fibroma. The ethmoidal location of the lesion may also be explained [5].

The cemento-ossifying fibroma was first observed in 35-year-old female with a long-standing large tumor in the mandible by Menzel in 1872. The peak incidence of cement-ossifying fibroma is in the third and fourth decades and is more prevalent in white racial groups. The lesion shows a marked predilection for the female with the female to male ratio being 2:1 and the mandible being most frequently involved around 70%. While significant percentage (22%) has been originate in the molar region of the maxilla, ethmoidal and orbital regions and exceptionally in the petrous bone [2]. The lesion is initially asymptomatic later the growth progresses to a noticeable swelling and mild deformity. Displacement of teeth may be an early clinical presentation. On rare occasions, dull pain or paraesthesia may be provoked due to pressure on an adjacent nerve. Expansion of buccal as well as lingual plates is supplementary with the larger lesions. Teeth in association with the lesion, seems to be vital sometimes as there is no associated root resorption observed [5, 6].

Radiographically, it is considered by three stages according to progression of the lesion: initial or early, mixed and mature stage. In the initial/early stage, the cemento-ossifying fibroma appears as a well-demarcated radiolucent lesion with no evidence of internal radio densities. As the tumor develops, there is evidence of calcification so that the radiolucent area becomes spotted with opacities until ultimately the lesion appears as lobulated complete radiopaque mass in the mature stage. An important diagnostic feature of cemento-ossifying fibroma is its centrifugal growth pattern rather than a linear growth, and therefore, the expansion of lesion takes place equally in all directions and present as a round tumor mass. The borders are well defined, and a thin radiolucent line signifying a fibrous capsule that separates the lesion from the surrounding bone. The cortical plate may be expanded, displaced and thinned still remains intact. Large aggressive mandibular lesions may cause a characteristic thinning and downward "bowing" of inferior border [7, 8]. Teeth adjacent to or involved in the lesion may be displaced but resorption is not associated with this tumor.

Histopathologically cemneto- ossifying fibroma exposes many delicate interlacing collagen fibers, infrequently arranged in discrete bundles, interspersed with large numbers of active, proliferating fibroblasts and cementoblasts sometimes osteoblasts may be seen [Figure 10]. Although mitotic figures may be present in few numbers, there is rarely any remarkable cellular pleomorphism is observed. As the lesion matures, the islands of cementum increase in number, enlarge later ultimately conjoin [9, 10].

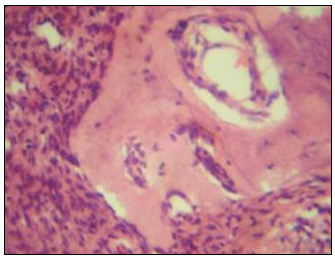

Fig 10: The postoperative view, the proliferating fibroblasts and cementoblasts sometimes osteoblasts

Cemento-ossifying fibroma is well circumscribed and separated from bone even if it has reached remarkable size, it should be excised conservatively, but complete resection of the lesion is a must. As the lesion is less vascularized and well circumscribed, there is relatively low risk in removal of the lesion from the surrounding bone.

Sakoda *et al*. described the procedure of a segmental resection of an extensive ossifying fibroma with the replacement of the excised segment after cryotherapy [8, 11]. Conservative surgery is therefore recommended even if the tumor is large with bowing and erosion of the inferior border of the mandible. If there is recurrence due to its aggressive nature the radical treatment such as an en bloc resection should only be considered. The prognosis is known to be fair and recurrence after surgical removal appears to be unusual.

Conclusion

As a conclusion, COF which may be encountered in various radiological and clinical features should be differentiated

from other pathologies by considering histopathological, radiological, and clinical features together. When considering the various clinical behaviors of the COF, purpose of the most appropriate treatment option and a long-term follow-up is mandatory because recurrences can develop for up to 10 years after treatment.

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