CEMENTO-OSSIFYING FIBROMA OF MANDIBLE: A CASE REPORT

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Cemento Ossifying fibroma is a benign fibro-osseous lesion that arises from the periodontal membrane. The periodontal membrane is a layer of fibrous connective tissue surrounding the roots of teeth. It contains multipotential cells that are capable of forming cementum, lamellar bone, and fibrous tissue. These tumours occur in the third and fourth decades of life, with predilection for women. The mandible is more commonly involved than the maxilla. This article describes a case of Cemento-ossifying fibroma in a 45-year-old female patient. A histopathological diagnosis of Cemento ossifying fibroma was made. An accurate diagnosis requires careful clinical, radiological and histological correlation in order to make an optimal treatment and an excellent outcome.

Key words: Cemento-ossifying fibroma, fibro-osseous lesions, gingiva.

Cemento-ossifying fibroma (COF) is a fibro-osseous lesion that arises from the periodontal membrane. [1]The periodontal membrane is a layer of fibrous connective tissue surrounding the roots of teeth. It contains multipotential cells that are capable of forming cementum, lamellar bone, and fibrous tissue.[2,3] Although central cemento-ossifying fibromas of the mandible are common, they are unusual in the maxilla.[4]

In 1872, Menzel gave the first description of a variant of ossifying fibroma, calling it a cemento-ossifying fibroma, in a 35-year-old woman with a long-standing large tumor of the mandible.[5]

This lesion has been referred to as osteofibroma, fibro-osteoma and benign fibroosseous lesion of periodontal ligament origin.[6] This lesion shares identical clinical, radiographic and histopathological features with a lesion previously termed as cementifying fibroma. In fact it is now widely accepted that these lesions are one and the same, prompting use of the term cemento ossifying fibroma by some. Support for a neoplastic etiology for ossifying fibroma includes examples of persistent, locally aggressive growth characteristics and the finding of recurrence in some cases. Chromosomal translocations have been described in a few cases of ossifying fibroma. In view of microscopic similarities with fibrous dysplasias and the cementoosseous dysplasias, some investigators regard the lesion as an example of a localized dysplastic process in which bone metabolism has been altered.

This lesion should be distinguished from fibrous dysplasia of bone and certain other fibro-osseous lesions that do not represent true neoplasia.[7] A close histogenetic relationship exists between the central cemento-ossifying fibroma and the central ossifying fibroma. It is based on the marked similarity between the two regarding predilection of age of occurrence, sex, race, location, roentgenographic appearance, and clinical behavior, these two lesions represent the same basic neoplastic process. The only difference between the two being in the type of cell involved and its end product-cementum in one case and bone in the other.

Clinical Features

The peak incidence of COF is the third and fourth decades and is more prevalent in white than black racial groups.[8] There is a marked predilection for the female sex, the female: male ratio being 5:1.[9,10] The mandible is more commonly involved than the maxilla. The lesion is generally asymptomatic until the growth produces a noticeable swelling and mild

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deformity; displacement of teeth may be an early clinical feature. On rare occasions, however, pain or paraesthesia may be elicited if pressure on an adjacent nerve ensues. Expansion of both buccal and lingual plates is associated with the larger lesions. Teeth in association with the lesion, retain their vitality and, as a rule, there is no associated root resorption.[11]

Radiographic Features

Radiographically, COF presents as a well-defined unilocular or multilocular lesion with smooth contours. The maturity of the lesion will determine the degree of radiopacity. The immature lesion may present as completely radiolucent, whereas the mature lesion may appear completely radio opaque. Displacement of adjacent teeth is common. One additional important diagnostic feature is that there is a centrifugal growth pattern rather than a linear one and therefore the lesions grow by expansion equally in all directions and present as a round tumor mass. Although, the tumour is of slow growing type, it may grow quite extensively and may even provoke mandibular fracture.[12]

Histopathological Examination

Histology reveals many delicate interlacing collagen fibers, seldom arranged in discrete bundles, interspersed with large numbers of active, proliferating fibroblasts and cementoblasts. As the lesion matures, the islands of cementum increase in number, enlarge, and ultimately coalesce. This, along with the increase in degree of calcification, accounts for the increase in the radio opaqueness of the lesions on the roentgenograms.

Treatment

Complete removal of the tumour mass, using enucleation or surgical resection is the treatment of choice. OF is usually well circumscribed and this facilitates its extirpation from the surrounding bone. In certain cases, bone grafting is essential to correct the defect. Radiotherapy has been proven ineffective and is contraindicated.[13] However, the treatment is governed by several factors, such as location, extent and size of the lesion. Recurrences are uncommon but it has been described.[9, 14]

Case Report

A 45-year-old female patient reported to the out patient department of Navodaya Dental College and Hospital, Raichur, Karnataka, India with a chief complaint of a painless swelling in the right posterior mandibular region for a period of 3 years. Initially, the swelling was small in size and showed a gradual increase to its present dimensions. The swelling extended from mandibular right first premolar region to the third molar posteriorly, and it obliterated the buccal vestibule. Her medical history was not contributory and physical examination disclosed no evidence of any systemic disease. No history of trauma to the mandible could be elicited.

Extraoral examination

Extra oral examination showed a diffuse swelling in the right mandibular region (Fig 1). The skin over the swelling was normal, and there was no local rise of temperature. On palpation the swelling had variable consistency. Buccal cortices were expanded. The lesion was smooth, nontender and hard; there was no accompanying cervical lymphadenopathy.



Fig. 1: Swelling in the right mandibular region causing facial esthetic deformity.

Intraoral examination

Physical examination revealed a large bony swelling extending from distal aspect of right sec-

ond molar to the right canine region with displacement of the external mandibular cortex. Intraoral examination showed a firm mass of size 4 X 5 cm with expan-sion of both mandibular walls and loss of integrity of the inner mandibular cortex extending from the position of the first premolar to the third molar region. The overlying mucosa was reddish pink in color and slightly swollen but there was no ulceration or fistula formation. On palpation, the swelling was bony hard in consistency but no tenderness or paraesthesia. There were no teeth in the right side of the mandible. Apart from poor oral hygiene, the rest of the oral cavity showed no unusual features (Fig 2).

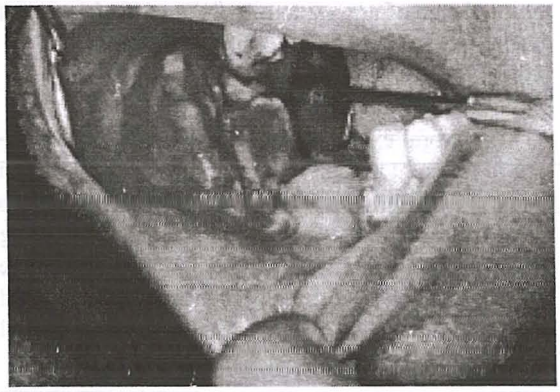


Fig. 2: Firm mass of size 5 X 4 cm extending from the first premolar to the third molar region.

Radiographic examination

Radiographically, orthopantomograph showed a large multilocular radioopaque lesion (approx 5x4 cm) that contained small foci of calcification extending from the first premolar region to the third molar region. The lesion was well demarcated with sclerotic border and heterogeneous in contrast. The lesion occupied the area corresponding to the clinical swelling i.e. extended mesiodistally from the area of first premolar to the position of the third molar area and superioinferiorly from the crest of the edentulous alveolar ridge to the area beyond the inferior alveolar canal, just above the inferior cortex of the mandible. The superior alveolar ridge is slightly elevated. No evidence of cortical erosion was noticed (Fig 3). The radiographic differential diagnosis included odontogenic myxoma, cemento-ossifying fibroma (COF), fibrous dysplasia(FD), focal cemento-osseous dysplasia (FCOD) and peripheral giant cell granuloma.

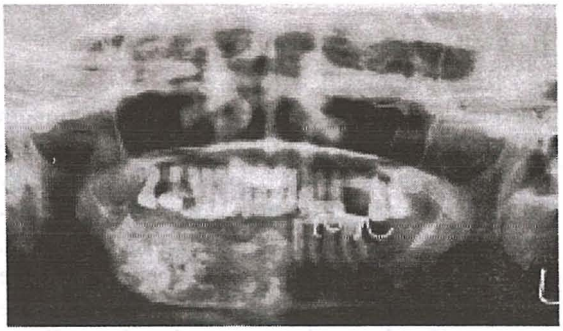


Fig. 3: OPG showed a large multilocular radio-opaque lesion (approx 5 X 4 cm) that contained small foci of calcification extending from the first premolar region to the third molar region.

A clinical diagnosis of Odontogenic Myxoma, Cemento Ossifying Fibroma (COF), Fibrous Dysplasia(FD), Focal Cemento Osseous Dysplasia (FCOD) and Peripheral Giant Cell Granuloma was made. The patient's medical history did not reveal any pathological condition. Therefore, surgical excision of the lesion was proposed to the patient.

Management

The surgical intervention was carried out in the Department of Oral and Maxillofacial Surgery. Wide surgical excision of the lesion was subsequently per-formed by means of a combined intra oral and extra oral approach. Segmental mandibulectomy was perfor-med from first premolar to third molar region. Reconstructiion was planned in the near future. The tissue was sent for histopathological evaluation.

Histopathologocal examination

Histopathological examination showed pseudoepitheliomatous hyperplasia of stratified

squamous epithelium. The deeper areas showed intense fibroblastic proliferation with calcifications of different sizes and shapes. Bony trabeculae rimmed by osteoblasts were seen (Fig 4).

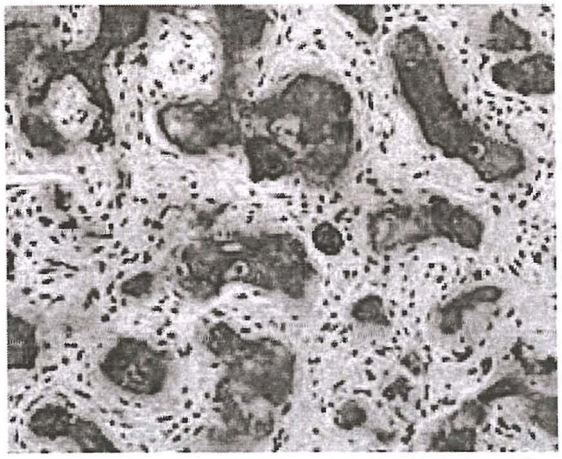


Fig. 4: Histopathologically deeper areas showed intense fibroblastic proliferation with calcifications of different sizes and shapes. Bony trabeculae rimmed by osteoblasts were seen.

The histologic, clinical and radiographic findings were consistent with cemento ossifying fibroma. The postoperative course was uncomplicated and there was no lesion recurrence up to one year of follow-up.

Discussion

Ossifying fibroma is classified as a cemento ossifying fibroma in the 1992 WHO classification of odontogenic tumors and is regarded as an osteogenic neoplasm.[15] It is also generally classified as a type of fibro-osseous lesion of the jaws. The cemento-ossifying fibroma is a fibro-osseous lesion that commonly affects third and fourth decades and is more prevalent in white than black racial groups.[8] There is a marked predilection for the female sex, the female: male ratio being 5:1.[9,10] The mandible is the common site, though the lesion may involve multiquadrants.[16] Cemento-ossifying fibroma arises from the periodontal membrane.[1] The periodontal membrane is a layer of fibrous connective tissue surrounding the roots of teeth. It contains multipotential cells that are capable of forming cementum, lamellar bone, and fibrous tissue.[2,3] In some cases, initial symptoms are present.[8] These are defined as demarcated, or rarely, encapsulated neoplasms that consist of fibrous tissue containing varying amounts of mineralized material that resembles bone and/or cementum.

A comprehensive review of the lesion was published by Eversole et al in their report on 64 cases.[17] Ossifying fibromas occur over a wide age range. The majority of cases are encountered in the third and fourth decades of life. There is a female predilection, with a female to male ratio of approximately 5:1. The mandible is more commonly involved than is the maxilla, and the premolar-molar region is the commonest site. Small lesions are often symptomless. Large lesions result in painless swelling of the involved bone which may cause obvious facial asymmetry. The radiographic appearance depends on the location and the amount of calcified tissue present. Varying degrees of radio-opacities and radiolucencies may be present. The fibroma tends to be well-demarcated. Large lesions often demonstrate a "downward bowing" of the inferior border of the mandible.[18]

Su and colleagues[8] reviewed clinical details of 75 cases of COF. The mean age was 32 years (range, 10-59 years). The authors reported that COF is not seen in patients over 60 years of age and is detected 10 years earlier than FCOD. Our patient has not consulted any dentist for the past three years before attending our OPD. It appears that the lesion was quite small at the initial presentation. COF may present as small radiolucent areas close to the apices of teeth that could be mistaken for periodontal pathology.[19] Nevertheless, the orthopantomograph view revealed that the lesion was more radio-opaque in appearance. Possibly, the tumour was undergoing maturation and ossification phase accounting for the increased density.[10] In addition, rapid followed by slow tumour growth may elucidate that the tumour was passing through a dormant phase.

Generally, COF characterised by a well-defined expansile bony mass and rarely associated with destruction or extraosseous soft tissue components.[10] Unlike the COF, the juvenile or aggressive (ACOFs) grow massively with extensive cortical expansion.[17, 20, 21] There are no histopathologic criteria that are predictive of aggressive behaviour or tendency for recurrence. Nevertheless, Zupi et al.[20] reported two features that may help in distinguishing ACOF from COF. Firstly, clinically, the ACOF occurs at a far lower mean age than the COF. Secondly, the histological pattern of the ACOF seems to be unique in being highly cellular with entrapped osteoblasts. However, considering the age, radiographic appearance and the growth behaviour, our case would clearly fit under the COF category, the non-aggressive type.

In fibrous dysplasia, an intimate continuity between the lesion and normal bone is generally found. The tumor expands throughout its length and the margins are diffuse and radiographically poorly defined. Its radiological structure is more homogenous than that of ossifying fibroma or cemento-ossifying fibroma, both of which are filled with radiopaque foci. Cemento-ossifying fibroma is a well-circumscribed tumor that grows expansively and has clearly defined margins. Lesions are oval, spherical, or multilocular and are clearly separated from the surrounding bone by osteolytic borders.[2]

Central cemento-ossifying fibromas are typically well-defined, solitary radiolucencies with scattered radiopaque foci. They maintain a spherical shape, expand cortical bone without causing perforation, and may cause tooth divergence.[21]

The neoplasm presents an extremely variable roentgenographic appearance, depending upon its stage of development. However, despite the stage of development, the lesion is always well circumscribed and well demarcated from surrounding bone, in contrast to true fibrous dysplasia. One additional important diagnostic feature of the lesion is its effect upon the inferior border of the mandible when the lesion reaches such a size as to encroach upon it. The central cementifying fibroma and its related lesions, the central ossifying fibroma and central cemento-ossifying fibroma, have a centrifugal growth pattern rather than a linear one. Therefore these lesions grow by expansion equally in all directions and present as round tumor masses. Other lesions (fibrous dysplasia) expand the cortex linearly and the outline of the expanded mandible is not in continuity with the remainder of the outline of the lesion.

Peripheral giant cell granuloma may also resemble the cemento-ossifying fibroma clinically, with both types of lesions commonly occurring anterior to the molars and presenting as pedunculated or sessile masses. However, peripheral giant cell granuloma can be ruled out on the basis of the size of the lesion (0.5-1.5 cm), its pedunculated nature, and the presence of peripheral cuffing and giant cells on histopathological examination. Peripheral cuffing, due to superficial erosion of the bone, is the pathognomic radiographic feature. Another lesion which also clinically resembles the cemento-ossifying fibroma is the calcifying epithelial odontogenic tumor. The age of occurrence of this tumor is 8-92 years with a mean age of 40 years. In calcifying epithelial odontogenic tumors, the common site of occurrence is the molar area, whereas the cemento-ossifying fibroma is generally seen anterior to the molars. Calcifying epithelial odontogenic tumor is generally associated with an unerupted or impacted tooth. Initial radiographic features of both the lesions are common (both being radiolucent) but as the lesion progresses it takes on a 'honeycomb' or 'driven snow' appearance. Histologically it can be differentiated by the presence of polyhedral epithelial cells as well as multinucleated giant cells.

Cemento-ossifying fibroma is a slow-growing lesion composed of cellular fibroblastic tissue containing basophilic masses of cementum-like tissue. In addition, varying amounts of bony trabeculae are interspersed within the lesion, giving it its characteristic features.[22] Reed[23] used the presence or absence of woven and lamellar bone in

histopathological section to differentiate the cemento-ossifying fibroma from the other fibro-osseous lesions. In uncomplicated cases, fibrous dysplasia contains no lamellar bone but, rather, has arrested woven bone. On the other hand cemento-ossifying fibroma and ossifying fibroma contain woven bone and are often rimmed by osteoblasts that have laid down layers of lamellar bone. Additionally cemento-ossifying fibroma may have areas of cementum, appearing as psammoma bodies embedded in a benign fibrous stroma.

The circumscribed nature of the lesion permits complete local enucleation or curettage of small lesions. Large lesions that have destroyed a considerable amount of bone may require segmental resection and reconstruction.

Mandibular reconstruction after resection is essential for the restoration of function and cosmesis. Otherwise, the patient will be crippled with a loss of significant function in mastication, the ability to swallow, and speech. An important part of the reconstruction is the use of removable or implant-supported dental prostheses.

Alloplastic and autogenous grafts or combinations of both are used widely in mandibular reconstruction. Bony reconstruction of the mandible requires the transplantation of viable bone from a suitable donar site to the remaining parts of the mandible. Current practice involves the transplantation of autogenous bone as the primary bone graft material. Three types of graft material are possible – corticocancellous blocks, free vascularised bone, and particulate cancellous bone.

Bone graft harvesting from the ilium for maxillofacial reconstruction is generally not associated with any significant morbidity. The use of corticocancellous block bone for jaw bone reconstruction has been reported extensively. However, in terms of the biology of bone grafting and healing, this may not be the most ideal method in the long term compared with the use of particulate cancellous bone because of the smaller numbers of marrow mesenchymal cells and viable osteoblasts present. This results in a deficiency of the replacement phase of the remodeling cycle and unpredictable resorption of bone. Further rehabilitation using dentures or implants is rendered impossible if significant resoption occurs.

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