



Original Article

Bilateral mixed density lesion of the mandible- A Rare case report with clinical, radiographical, biochemical and histopathological findings.

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ABSTRACT

Cemento-osseous dysplasias are a group of disorders known to originate from periodontal ligament tissues and involve, essentially, the same pathological process. They are classified depending on their extent and radiographic appearances, into three main groups: periapical (surrounds the periapical region of teeth and are bilateral), florid (sclerotic symmetrical masses) and focal (single lesion) cemental dysplasias. Florid cemento-osseous dysplasia (FCOD) is a type of fibro-osseous lesion and represents a reactive process in which normal bone is replaced by poorly cellularized cementum-like materials and cellular fibrous connective tissues. It is strictly localized to the tooth-bearing or edentulous areas, often occurring bilaterally with symmetric involvements. FCOD has also been classified as gigantiform cementoma, chronic sclerosing osteomyelitis, sclerosing osteitis, multiple stenosis and sclerotic cemental masses. Radiographically, it appears as a dense, lobulated masses, often symmetrically located in various regions of the jaws and patients do not have laboratory evidence. Cone beam computed tomography, because of its ability to give axial, sagittal, and frontal views, is useful in the evaluation of these lesions. The best management for the asymptomatic FCOD patient consists of regular recall examinations with prophylaxis. This article reports the case of a middle aged female patient who was diagnosed with FCOD on the basis of clinical, radiographic, biochemical and histological findings.

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Introduction

They are more commonly seen in middle-aged black women (in some series, more than 90 percent of patients), although it also may occur in Caucasians and Asians. (3) The reason for this racial and gender predilection is unknown. (4) In some cases, a familial tendency has been reported, but most examples appear to represent isolated instances. (5) The lesions show a marked tendency for bilateral and often quite symmetric involvement, and it is not unusual to encounter extensive involvement of all four posterior quadrants. The process may be totally asymptomatic and, in such cases, the lesion is detected when radiographs are taken for some other purposes. Symptoms such as dull pain or drainage are almost always associated with exposure of sclerotic calcified masses in the oral cavity. This may occur as the result of progressive alveolar atrophy under a denture or after extraction of teeth in the affected area. (6)

Radiographically, the lesions appear as multiple sclerotic masses, located in two or more quadrants, usually in the tooth-bearing regions. They are often confined within the alveolar bone. Computed tomography (CT), because of its ability to give axial, sagittal, and frontal views, is useful in the evaluation of these lesions. (7)

Histologically, these lesions are composed of anastomosing bone trabeculae and layers of cementum-like calcifications embedded in a fibroblastic background. Management of FCOD's may be difficult and not very satisfactory. The disease may persist for indefinite periods of time being asymptomatic. For the asymptomatic patient, the best management consists of regular recall examinations with prophylaxis and reinforcement of good oral hygiene. (6,7)

Since fibro-osseous lesions tend to have a wide range of presenting clinical and radiographic appearances, the aim of this case report is to show that it is imperative for the dentist to be familiar with the different presentations of these benign lesions in order to establish an appropriate diagnosis and treatment planning.

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Case Description:

A 35 year old female, reported to the out-patient of oral medicine and radiology department, with a chief complaint of deposits and missing tooth at right and left lower back tooth region since 7 years. Patient was apparently healthy seven years back after which patient starting eliciting continuous sharp pain at her right and left back tooth and got them removed with no post-operative complications. Till date, the patient does not complain of any associated pain or discomfort in the missing tooth region. No history of recent trauma. Patient gives a history of Hypothyroidism since 10 years and patient is on Tab. Eltroxin 50mcg. Patient brushes once daily and consumes mixed diet. No relevant family history was elicited. On, General examination, the patient was well built, well nourished, calm, conscious, cooperative, well oriented to time and space with normal gait, with no abnormal findings in vital and constitutional signs. No abnormalities were detected on extraoral examination. (Fig 1) On intraoral examination, mild stains and calculus was noted, with missing 36 and 46. Based on the history and clinical examination, a provisional diagnosis of Generalized chronic gingivitis and Partially edentulous, was made. On patient's consent for an implant treatment, an OPG was taken. (Fig 2) Incidentally, bilateral, mixed density lesion, roughly measuring around 2cm×2cm was noted within the alveolar bone in relation to 36 and 46, above the level of inferior alveolar canal, with surrounding radiolucency closer to alveolar crest. The periphery of the conglomerated mass was well defined and regular. The internal structure was mixed radiopaque-radiolucent. The content of the lesion was largely radiopaque with irregular mass of calcified tissue. For a more detailed assessment, a Cone beam computed tomography (CBCT) was taken after patient's consent. (Fig3)

CBCT revealed bilaterally well defined mixed density lesion with radiopaque predominance measuring approximately 30 mm× 24 mm in the periapical region of right and left posterior mandible at the region of 36 and 46, extending anteriorly upto the premolar and posteriorly upto the second molar, superiorly upto the alveolar crest of the mandible and inferiorly few mm above the mandibular canal. (Fig 4, 5) Axial sections of 1 mm thickness were obtained. Mild expansion of buccal and lingual cortical plates was evident bilaterally with no evidence of cortical perforation. (Fig 6a, 6b) Superior border of mandibular canal was intact. (Fig 5) Radiographic differential diagnosis included, Cemento ossifying fibroma, florid cementosseous dysplasia, complex odontomas, diffuse sclerosing osteomyelitis, idiopathic osteosclerosis and calcifying epithelial odontogenic tumour. Biochemical analysis of serum alkaline phosphatase, calcium and phosphorus was carried out to differentiate from Paget's disease and was found within normal limits.

A small incisional biopsy was done on the left side under LA. Histopathological findings revealed formations of dense sclerotic calcified cementum-like masses. (Fig 7) The lesion was composed of cementum like substances characterized by islands of calcified deposits and areas of loose fibro-collagenous stroma, which showed the evidence of proliferation. There are thick, confluent curvilinear trabeculae with little fibrotic stroma. Periphery of the lesion showed globular or ovoid structures of cementoid appearance involved by

thin fibrous tissue. The cementum-like substances mainly showed cellular structure, all features suggesting of florid cement-osseous dysplasia. Bilaterally, an excisional biopsy was not done due to the avascular nature of the lesion which contributes to the susceptibility of the lesion to severe infection, bone sequestration and osteomyelitis when an extensive surgery is performed. The treatment plan included good oral hygiene maintenance and periodic follow-up.

FIGURES AND FIGURE CAPTIONS

Fig 1- Profile: No facial asymmetry evident



Fig 2- Panoramic view: Bilateral, mixed density lesion, roughly measuring around 2cm×2cm, within the alveolar bone in relation to 36 and 46, above the level of inferior alveolar canal, with well defined periphery and a largely radiopaque internal structure.

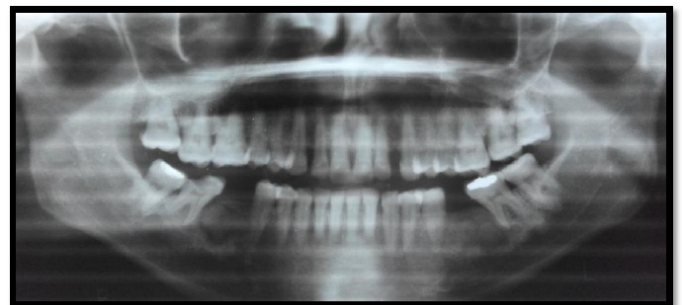


Fig 3- CBCT Panoramic view: Evidence of multiple mixed density lesions bilaterally in posterior mandible in relation to the peri-apical region of 36 and 46.



Fig 4- CBCT Sagittal view: Depicting mesio-distal extension of the lesion. High density mass surrounded by a well defined peripheral low density region.



Fig 5- CBCT Coronal section: Showing superior-inferior extension of the lesion. The lesion do not appear to be in contact with mandibular canal.



Fig 6 (a, b) CBCT best fit section: Evidence of mild bucco-lingual expansion and no evidence of cortical perforation. Irregular high density mass surrounded by a well defined peripheral low density region.

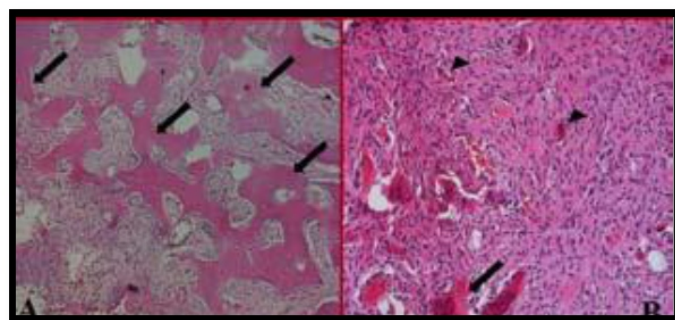


(a)



(b)

Fig 7- Histopathological picture: The lesion showed woven bone trabeculae (arrow) and cementum-like mineralization (arrowhead) within cellular fibrous connective tissue (Hematoxylin-Eosin A:x100, B:x200).



Discussion

FCOD has been described as having three developmental stages, each with specific radiographic features. In the early or osteolytic stage, radiographs show a well-defined radiolucent lesion. In the intermediate stage, it displays a mixture of radiolucent and radio opaque architecture. The last mature stage is characterized by a definite radiopacity, present in the major part of the lesion. (8) CBCT can be used in differentiation of FCOD from other lesions which have similar sclerotic appearance on conventional radiographs because it gives the clear view of axial aspect of the lesion. In our case, the conventional radiograph showed a mixed density lesion with distinct borders. By using a CBCT, the state of the lesion relative to the buccal and lingual cortical plates could be assessed, which might not be possible on the conventional radiographs. CBCT axial cuts showed the mild expansion of the buccal and the lingual plates bilaterally, with no evidence of cortical perforation. (Fig 6a,6b) Also, the inferior alveolar canal was not involved with the lesion. (Fig 5) To the best of our knowledge, a few FCOD cases diagnosed with CBCT have been reported in the literature; furthermore, CBCT images are more useful diagnostic tools for identifying the location and extent of the lesion with less radiation while compared to a computed tomography. (9)

Waldron stated that "in the absence of good clinical and radiologic information, a pathologist can only state that a given biopsy is consistent with a fibro-osseous lesion" and that "with adequate clinical and radiologic information, most lesions can be assigned with reasonable certainty into one of several categories". (10)

Histopathologically, FCOD is a benign fibro-osseous lesion that must be differentiated from other benign fibroosseous lesions such as fibrous dysplasia, ossifying fibroma, Paget's disease, and sclerosing osteomyelitis on the basis of combined clinical, radiographic, and histological features. However, FCOD do not appear to be developmental in nature such as fibrous dysplasia (11), nor do it show the characteristics of neoplasia such as ossifying fibroma. (12, 13) Paget's disease of the bone may have a cotton wool appearance. Whereas florid cemento-osseous dysplasia is centered above the inferior alveolar canal and its cervical two thirds are normal. (14) Paget's disease is often polyostotic, involving other

bones such as spine, femur, skull, pelvis and sternum (15) and produces biochemical serum changes, such as elevated alkaline phosphate levels. (16) No biochemical alterations and others bone involvement were found in our case.

Chronic diffuse sclerosing osteomyelitis is a primary inflammatory condition of the mandible presenting with cyclic episodes of unilateral pain and swelling and shows a single area of diffuse sclerosis containing small, ill-defined osteolytic areas. whereas florid cemento-osseous dysplasia is seen as multiple round or lobulated opaque masses. Chronic diffuse sclerosing osteomyelitis involves the body of the mandible from the alveolus to the inferior border and may extend into the ramus. Florid cemento-osseous dysplasia has been interpreted as a dysplastic lesion or developmental anomaly arising in tooth-bearing areas. In addition, florid cemento-osseous dysplasia is frequently associated with black women, while chronic diffuse sclerosing osteomyelitis is seen predominantly in adult Caucasian men. (17, 18) Florid cemento-osseous dysplasia may have similarities with jaw bone changes in familial adenomatosis coli (Gardner's syndrome) (19), but florid cemento-osseous dysplasia has no other skeletal changes or skin tumours or even the dental anomalies that are seen in this syndrome. Florid cemento-osseous dysplasia may be familial with an autosomal dominant inheritance pattern, but there are only a few examples in the literature in which the familial pattern has been confirmed. (20) In the present case no familial aspects of the disease could be established.

Osteoid osteoma and osteoblastoma occur during the second decade of life. The dull and nocturnal pain that FCOD lacks is the major symptom associated with osteoid osteoma and osteoblastoma. (21) Paget's disease had a site preponderance for the maxilla and can be ruled out after a blood examination demonstrating the high serum alkaline phosphate level, which is within normal limit in patients with FCOD. (21,22)

The present case may resemble chronic sclerosing osteomyelitis but there is no clinical history of pain or pus drainage in relation to the lesion site. (23) So we ruled out such differential diagnosis. The present case shows the cemento-osseous tissue with absence of fusion to the apex of the tooth root, which rules out the possibility of cementoblastoma. (24)

It is important to differentiate lesions of COD from a true neoplasm, Ossifying Fibroma (OF). A periodic follow-up is recommended in order to ensure that no further enlargement or expansion occurs, features that argue in favour of an ossifying fibroma that was merely encountered in an early stage of development. (23) This differential diagnosis is also ruled out because in our case, at a 3 year follow-up, the lesion didn't encounter any modification.

In our case, there was mild buccal and lingual cortical plates expansion. Melrose et al., (25) reported jaw expansion with intact thinned cortices in their series of 34 florid COD. Because of the dense bone with poor vasculature found in FCOD, the periapical infection may progress into osteomyelitis. Therefore, the patient was motivated about the oral hygiene, referred to Prosthodontics Clinic to rehabilitate her edentulousness. She has been followed up over the last 3 years and FCOD has remained asymptomatic.

Conclusion

Normally, a pathologic lesion in the jaw requires a diagnosis based on clinical presentation, epidemiology, radiographic features and histopathologic findings. Patients with FCOD lesions should undergo clinical and radiographic follow-up for several years to ensure no destructive changes in the jaw. It is of major importance that dentists should be very careful in such cases because this kind of mixed density lesions may be misdiagnosed or overlooked and this may cause unnecessary endodontic or surgical treatments that may lead to complications. Learning from this case, we suggest that when a radiolucent or mixed lesion is found incidentally, a detailed 3D radiographic study may help to differentiate an inflammatory lesion from a cement-osseous dysplastic lesion. Therefore, this present case highlights the necessity and the role of oral and maxillofacial radiologist to make the differential diagnosis, an important and an essential one in doubtful cases.

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