CASE REPORT

Florid Osseous Dysplasia in Edentulous Maxilla and Mandible: A Rare Case Report.

Latha Mary Cherian, Krishnapriya S, Indu M, Kristina Sabu, Swathi Sanil, Manjusha Mohanan

Abstract

Introduction: Florid osseous dysplasia (FOD) has been described as a reactive fibro-osseous lesion affecting the jaw bones especially the tooth bearing areas. Usually, this lesion is asymptomatic unless accidentally detected during radiographic examination for any other purpose. Although the etiology is unclear, several theories suggests that its origin was related to reactive or dysplastic changes within the periodontal ligament.

Case Presentation: Presenting a case of Florid Osseous Dysplasia within the edentulous maxilla and mandible of 65-year-old female patient. Patient reported with a diffuse swelling in the edentulous maxillary and mandibular posterior regions intraorally. Radiographically, multiple radiopaque lesions were noted bilaterally in the posterior mandible and also in the right posterior side of the maxilla. On microscopic examination, the lesion was composed of spicules of bony trabeculae with osteocytes in lacunae and intervening fibro-fatty connective tissue with plump fibroblasts.

Management and prognosis: After the incisional biopsy, surgical recontouring was done along with further regular follow up and observation.

Conclusion: Diagnosis of the jaw lesions is based on clinical findings, radiographic features, and histological picture, whereas FOD can be diagnosed based on its clinical and radiographical features. Even though the surgical approach may lead to many complications like osteomyelitis, infection, fracture etc., a biopsy may be done to confirm the diagnosis.

Keywords: Cemento- osseous dysplasia, Fibro osseous lesions, Florid Osseous dysplasia, Osteomyelitis, Surgical recontouring.

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Introduction

Florid osseous dysplasia (FOD) has been classified as a reactive (dysplastic) lesion arising in the tooth- bearing area.[1] As a benign fibro-osseous lesion, FOD can be referred to as a group of osseous lesions with multi quadrant involvement.[2] The term FOD has been used to imply rather broad parameters of an exuberant variant of osseous dysplasia, defined by Robinson as an abnormal reaction of bone to irritation or stimulation.[3] It is synonymous with Florid Cemento Osseous Dysplasia (FLCOD).[4] This reasonably well-defined entity is usually seen in the jawbones of middle-aged women with an unknown etiology. One theory proposes that FOD could be caused by defective bone remodeling triggered by local injury or, possibly, an underlying hormonal imbalance.[1] Sometimes this lesion may be associated with extraction sites, supporting the etiology of an abnormal bony reaction to injury or trauma. There is no clear evidence for gender and racial predilection. Few radiologic and histopathologic studies showed that the lesion is limited to the tooth-bearing areas of the jaws. Some cases have been shown a familial distribution.[5] Clinically it is asymptomatic and detected accidentally during radiographic examination for any other purposes. When this lesion gets infected, symptoms like dull pain, drainage, exposure of the lesion in the oral cavity, focal expansion, and facial deformities can be seen.[1]

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In this case report, we present a case of non-familial Florid Osseous Dysplasia in a 65-year-old Indian female with a completely edentulous maxilla and mandible.

Case Report

A 65-year-old female reported with a complaint of swelling and pain in the right back region of her cheek for several months. The patient had a history of swelling in the posterior region of the mandible (38, 48 regions) and had previously undergone surgical management as well as full mouth extraction 5 years prior.

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Further medical records pertaining to the surgical procedure and histopathological diagnosis were not available. No relevant family history or social history is noted. Intra orally, a completely edentulous maxilla and mandible with a flabby ridge was observed in the 47 and 48 area. The patient had been wearing a complete denture for the previous 4-5 years. On intra oral examination, a diffuse swelling was present in the maxillary and mandibular posterior regions, i.e., 18, and 48 regions respectively (Fig. 1). On palpation, the swelling was tender and hard in consistency. An orthopantomograph (OPG) revealed multiple radiopaque lesions bilaterally in the posterior mandible and also in the right posterior side of the maxilla (Fig. 2). A differential diagnosis could include chronic diffuse sclerosing osteomyelitis, or Pagets disease. An incisional biopsy was taken from the right body of the mandible and the right posterior maxilla which was sent for histopathological examination.

Hematoxylin and eosin stained sections of multiple hard tissue bit from the mandibular region reveals dense lamellated bone containing osteocytes in lacuna. (Fig. 3). Resting and reversal lines were present. The intervening stroma was hypercellular with plump fibroblasts. Occasional osteoblastic rimming can be seen. Similarly, a histopathologic section of a maxillary specimen reveals spicules of bony trabeculae with osteocytes in lacunae. Resting and reversal lines were seen with occasional osteoblastic rimming. Fibro-fatty connective tissue was seen interspersed between bony trabeculae. (Fig 4). The histopathological picture was suggestive of fibro-osseous dysplasia. Thus, the final diagnosis of florid osseous dysplasia was confirmed, correlating with the clinical and radiographic features.

Discussion

The World Health Organization (2017) fourth edition of the Classification of Head and Neck Tumours, introduced "fibro-osseous lesion" (FOL) as a lesion under a group for the first time in the WHO classification of odontogenic and maxillofacial bone tumors. This category consists primarily of three lesions: fibrous dysplasia (FD), cemento-ossifying fibroma (COF), and cemento-osseous dysplasia (COD). The WHO 2017 edition changed the name of osseous dysplasia to another term, cemento-osseous dysplasia. CODs are the most frequently occurring FOLs. Cemento-osseous dysplasia (COD) is classified into three types based on clinical characteristics, location, and radiographic features: focal cemento-osseous dysplasia, florid cemento-osseous dysplasia, and periapical cemento-osseous dysplasia. (Table 1)[6] Both the focal and florid subtypes are the most common types found in middle-aged to elderly females of sub-Saharan African and East Asian origins. A review of the literature revealed that only about 2% of cases in the Indian population have been documented. It was also noticed that cases involving edentulous jaws had not been reported.[10,16] Macdonald-Jankowski compared the prevalence of FCOD across races and found that 59.6% of black women, 37.2% of oriental women, and 3.2% Indian or Cauca-

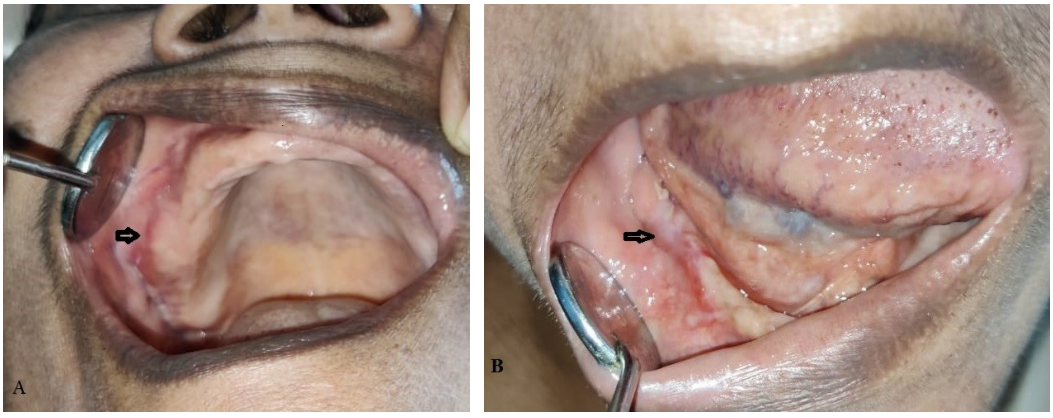

Fig. 1 A & B: Shows a diffuse swelling in the edentulous 18 region (A) and 48 region (B).

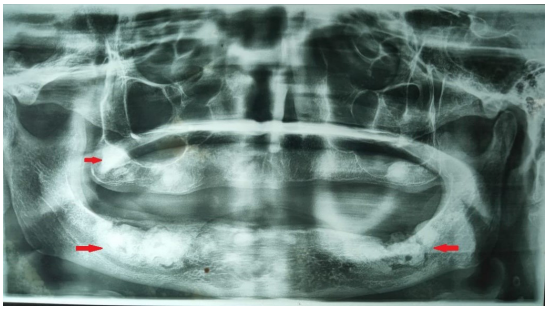

Fig. 2: Multiple radiopaque lesions bilaterally in the posterior mandible and also in the right posterior side of the maxilla.

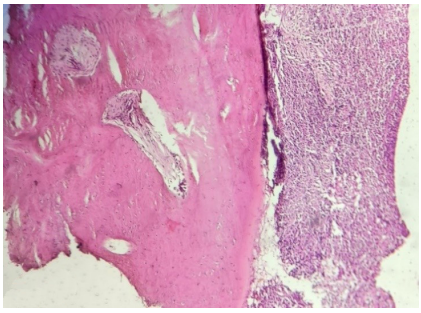

Fig. 3: Dense lamellated bone with osteocytes within the lacunae and hypercellular connective tissue stroma with plump fibroblasts (H&E 10x)

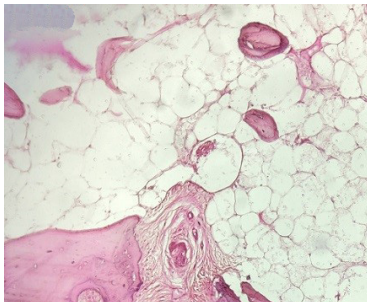

Fig. 4: Lamellated bony spicules with intervening fibro fatty connective tissue stroma (H&E 10x)

sian people had the lesion.[7] Periapical cemental dysplasia is usually seen at the apices of anterior teeth and is mostly of smaller size than 1 cm. Focal cemeto-osseous dysplasia is characterised by the presence of two or more mandibular anterior tooth regions or the apices of the molar regions. FLCOD appears bilateral, often quite symmetrical, and rarely grows more than 2 cm.[8]

Table 1: Classification of Fibro-osseous lesions WHO 2017

Fibro-osseous lesions
A. Fibrous dysplasia
B. Ossifying fibroma- 1. Cemento- ossifying fibroma 2. Juvenile ossifying fibroma a) Psammomatoid juvenile ossifying fibroma b) Trabecular juvenile ossifying fibroma
C. Osseous dysplasia 1. Familial Gigantiform Cementoma 2. Cemento- osseous Dysplasia a) Periapical cemento-osseous dysplasia b) Focal cemento-osseous dysplasia c) Florid cemento-osseous dysplasia

The term "florid" refers to its excessive and widespread location.[9] M. Benaessa suggested that COD mainly affected the mandible (62.4%), followed by involvement of both the maxilla and the mandible (24.5%), and maxilla (13.1%). According to his studies, among the patients with known COD subtypes, florid COD predominated (65%).[4]

In 1976, Melrose et al. suggested the term "florid osseous dysplasia" to describe a condition characterised by exuberant multiquadrant masses of cementum and/or bone in both jaws, and in some cases, simple bone cavity-like lesions in affected quadrants.[10] FLOCD exhibits multifocal involvement which is not limited to the posterior mandible. Although many cases affect only the posterior portions of the jaws, synchronous involvement of the anterior mandible may be observed as well. The lesions show a tendency for bilateral and fairly symmetrical involvement of the mandible, and occasionally there may be extensive involvement in all four quadrants.[11] FLCODs are not associated with any other extragnathic abnormalities.

Literature search has been found that FLCOD had three developmental stages with different radiographic images as given below. Well-defined radiolucent areas with loss of lamina dura and periodontal ligament are the radiographic features of the first or osteolytic stage. Within the second or cementoblastic stage, small radiopacities appear within the radiolucent area because of the deposition of cementumlike droplets in fibrous tissue. In the last stage, definite radiopacity was present in the majority of the lesions.[12] Multiple radiopaque masses are seen radiographically in this case as well, which may correspond to the third or final developmental stage.

The aetiology of the disease is unclear. Waldron et al[1] proposed that reactive or dysplastic changes within the periodontal ligament could be a cause of the disease. Similarly, Melrose et al.[10] suggested that disorderly bone production might result in obstructed drainage of interstitial fluid and thus lead to cyst or cavity formation. These lesions are characterised by the replacement of bone by connective tissue matrix displaying varying degrees of mineralization within the type of woven bone or cementum ¬like round basophilic acellular structures.

According to Ackerann and Atlini, the clinical features of osseous dysplasia are characteristics of the different clinicopathologic variants such as solitary, multiple, florid, and periapical.[13] The classic feature of FLCOD is symmetric bony hard swelling of the jaws, often involving all four quadrants. The masses may become large and cause considerable facial deformity. Symptoms such as pain or drainage are mostly associated with exposure of the sclerosing calcified masses in the oral cavity. Our patient had symmetrical bony hard swelling in all four quadrants especially in the molar area, as well as pain, which was identical to the classic feature of osseous dysplasia.

The biochemical findings of the patients with FLCOD is aessentially normal.[11] Familial FCOD is quite uncommon, and only a few cases have been reported in the literature where the hereditary nature of the lesion can be demonstrated. The familial form tends to occur in younger individuals and is characterised by more expansile lesions that may recur after surgery. In all of the familial cases reported in the literature, FCOD appears to be inherited as an autosomal dominant trait with variable phenotypic expression.[14] Srivastava et al. reported a case of familial FLCOD associated with multiple impacted teeth.

This was a non-familial lesion associated with completely edentulous maxilla and mandible.

The radiographic image shows lobular radiopacities that grow with lesion maturation and are surrounded by radiolucent areas. Most of the cases were found in the mandibular premolar molar region. The classic appearance includes diffuse, lobular, irregular shaped radiopacities throughout the alveolar process, which occur above the mandibular canal and are limited to the tooth-bearing area. Also, the lesions demonstrate a maturation pattern similar to that in other forms of cemento-osseous dysplasia.[4,11]

Initially, the lesions are predominantly radiolucent/osteolytic stage but with time become mixed (radiolucent and radiopaque), then predominantly radiopaque with only a thin radiolucent rim (osteogenic stage). On occasion, a lesion can become almost totally radiopaque and blend with the adjacent normal appearing bone. Typically, the radiopacities remain separated from adjacent teeth with an intervening, intact periodontal ligament space. In some end stage lesions, the cemento-osseous material may fuse with the tooth root surface to produce thickened root apices surrounded by radiolucency (or a "hypercementosis-like appearance"). Both dentulous and edentulous areas may be affected, and involvement appears to be unrelated to the presence or absence of teeth.[15] Multiple radiopaque lesions were found in the edentulous mandible and maxilla in this case, whereas the majority of the cases reported were in dentulous jaws.

In some cases of FLCODs, computed tomography, particularly axial CT (Ariji Y et al) and three-dimensional imaging, has been described as useful.

Histopathologic features of FLOCD are typically fragments of cellular fibrovascular connective tissue with scattered haemorrhagic areas and a variable mixture of woven bone, lamellar bone, and cementum like particles. As the lesion matures, the ratio of fibrous connective tissue to mineralized material decreases. Bony trabeculae become thick and curvilinear, with shapes similar to ginger roots. In the final radiopaque stage, the individual trabeculae fuse to form sheet like or globular masses of sclerotic, disorganised cemento-osseous material.[11,15] Here, in our case, histopathologically the lesion shows dense lamellated bone containing osteocytes in

lacunae with intervening stroma.

Immunohistochemical studies of FLCODs and other FOLs of the jaw are rare in the literature. Burkhardt conducted a study in fibro-osseous cemental lesions of the jaw using vimentin, alpha1-antitrypsin, lysozyme, and S 100 protein markers and found a positive reaction to vimentin.[12]

The differential diagnosis of FLCOD includes Paget's disease of bone, chronic diffuse osteomyelitis, Gardner's syndrome, and familial gigantiform cementoma. Paget's disease of bone may mimic FLCOD on radiological evaluations. Paget's disease involves the entire mandible and exhibits loss of lamina dura, whereas FLCOD is seen above the inferior alveolar canal.[15] In our current case, the radiographic appearance does not involve the entire mandible, but at the same time, the maxilla is also involved. Therefore, Paget's disease can be ruled out.

Cemento-ossifying fibroma is a neoplastic lesion, that shows more severe buccolingual expansion than FLCOD since it is not neoplastic. Commonly, FCOD is not seen with skin tumors, dental anomalies, and skeletal changes like Gardner's Syndrome, familial adentomatosis coli, but may have similarities with jaw bone changes. CEOT, ossifying fibroma, and fibrous dysplasia are benign, slow-growing expansile lesions, while FLCOD rarely shows bone expansion.

As most of the FOD lesions are asymptomatic, diagnosis of such disease is predicated on the basis of clinical and radiographic findings. If the radiological and clinical findings are enough to diagnose, biopsy should be avoided because of risks of significant post-operative infection.

The surgical approach may lead to increase the risk of osteomyelitis of the bone due to the lack of vascularity of the lesion and, complete removal of necrotic tissue may result in a large discontinuity defect which ultimately results in patient morbidity. Hence, in asymptomatic patients, the recommended treatment is observation and regular radiographic follow-up. Surgical recontouring should be the treatment of choice in cases where there is only cortical expansion with or without mucosal perforation. In symptomatic cases, administration of antibiotics is indicated, but sometimes it may not respond to antibiotics due to the avascular nature of the lesion, requiring surgical debridement and enucleation.[11] In this present case, surgical recontouring was done along with further regular follow up and observation.

Conclusion

Florid osseous dysplasia is a benign, multifocal fibro-osseous dysplastic process of tooth-bearing regions. Most cases of florid cemento osseous dysplasia are asymptomatic and are found during a routine radiographic examination. Diagnosis of the jaw lesions is based on clinical findings, radiographic features, and histological picture, whereas FLCOD can be diagnosed based on its clinical and radiographical features. Even though the surgical approach may lead to many complications like osteomyelitis, increased risk of infection, fracture of the jaw etc., a biopsy may be done to confirm the diagnosis. CBCT images are also useful for identifying the location and extent of the lesion and, moreover, for diagnosing it. Uncomplicated FCOD may remain asymptomatic for an indefinite period of time and requires regular follow up and no invasive therapy.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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