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A Case Report & Review of Literature of Florid Cemento-Osseous Dysplasia (FCOD)

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Florid cemento-osseous dysplasia (FCOD) is a rare, multifocal fibro-osseous dysplasia affecting tooth-bearing areas of the jaw, characterized by replacement of normal trabecular bone with disorganized osseous tissue and dense acellular cementum in a fibrous stroma. It is one of the variants of non-neoplastic fibro-osseous lesions known as cemento-osseous dysplasias (CODs), thought to arise from elements of the periodontal ligament. The word 'florid' relates to extensive, wide spread presentation of the disease in the jaws. FCOD is most commonly found in middle-aged, black women with the prevalence of 5.5%. It is even rarer based on the reports Indian

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Population. It predominantly affects middle-aged women of African descent, less frequently in Caucasian and Asian groups. Generally florid cemento-osseous dysplasia is asymptomatic, and is usually detected during radiological examination. In this report, we present a case of a 46-year-old female patient diagnosed with diffuse sclerosing osteomyelitis later on histopathological examination revealed FCOD.

Keywords: FCOD; fibrous stroma; orthopantomogram; craniofacial skeleton.

1. INTRODUCTION

"Floridcemento-osseous dysplasia (FCOD) is a"fibro-osseouslesion" that characteristically affects the jaw bones of the middle-aged with multi-quadrant radiopaque cementum-like masses. In thepast, the condition was known with a variety of names causing confusion in diagnosis and treatment" [1]. "The condition is usually asymptomatic and needs no treatment as such. These lesions are clinically asymptomatic and may be found as incidental radiological finding presenting as multiple radiopaque masses within peripheral radiolucent rim located in two or more quadrants usually in tooth-bearing areas" [2].

2. CASE REPORT

A 46-year old female patient reported to the outpatient department with a chief complaint of pain and swelling on left lower back tooth region for 20 days. The patient was hypothyroid for 2 years and under medication. No history of drug allergy or any relevant family history was seen. No deleterious habits were present. General examination revealed her to be well built and healthy. On extraoral examination, no asymmetry was detected. On intraoral examination, a solitary oval swelling on the left lower back tooth region was noticed in relation to the first molar region. The overlying skin was intact with no surface discoloration or rise in local temperature. On palpation, it was hard and non-tender. Further CT and orthopantomogram were advised to diagnose the lesion. It revealed ill defined radiolucency involving right and left quadrants. On orthopantomogram, ill-defined radiolucencies at the apices of 31, 32, 33, 41&42 surrounded by thick sclerotic borders was evident. Defined radiolucency surrounded by thin radiopaque rim was noticed irt 36 region. Conglomerate radio-opaque masses were attached to mesial root of 37. Mixed radiolucent and radio opacity along with root resorption was seen in apical region of 46, 47. Thick curvilinear trabecular pattern was noticed in relation to 38 & 48 regions (Fig. 1). Axial CT revealed moderate bony expansion in posterior region of mandible irt 46 region along with thinning of buccal cortical plate (Fig. 2A) as well as irt 36 region (Fig. 2B) and mild expansion in anterior region of the mandible (Fig. 2C). On the basis of case history, clinical presentation and radiographic features, a working diagnosis of diffuse sclerosing osteomyelitis was made.

For confirmatory diagnosis, under all aseptic precautions, Incisional biopsy under LA was performed. The specimens of hard and soft tissue bits were sent for histopathological examination. The H&E-stained soft tissue sections from left mandibular region (36 tooth region) showed fibrocellular connective tissue with numerous angular, oval and stellate shaped cells resembling undifferentiated mesenchymal cells in a basophilic background and numerous fibroblasts with inconspicuous cell borders and irregularly arranged collagen fibers. Few osteoid like areas with osteocytes within the lacunae along with round to oval cementum like spherules were evident (Figs. 3, 4). Tissue bits from 46, 47 tooth regions showed fibrocellular connective tissue with numerous angular, oval and stellate shaped cells, collagen fibres, osteoid like areas (Fig. 5) and numerous hemorrhagic areas with mixed inflammatory infiltrate predominant of lymphocytes, plasma cells and neutrophils were evident. Then the area was thoroughly cleaned with curette and the adjacent bony areas were sent for histopathological examination that revealed similar features as incisional biopsy.

Hence, correlating the clinical, radiographic and histopathological features florid cemento-osseous dysplasia (FCOD) was formulated as the final diagnosis. Patient is under regular clinico-radiological follow-up.

Differential diagnosis: There are various lesions in the jaws that have a similar radiographic appearance like periapical infection, chronic diffuse sclerosing osteomyelitis, polyostotic fibrous dysplasia.

Fig. 1. OPG revealed defined radiolucency surrounded by a thin radiopaque rim along with conglomerate radio-opaque masses on the left side of the mandible. Mixed radiolucent and radio opacity with ill defined borders on right side of the mandible

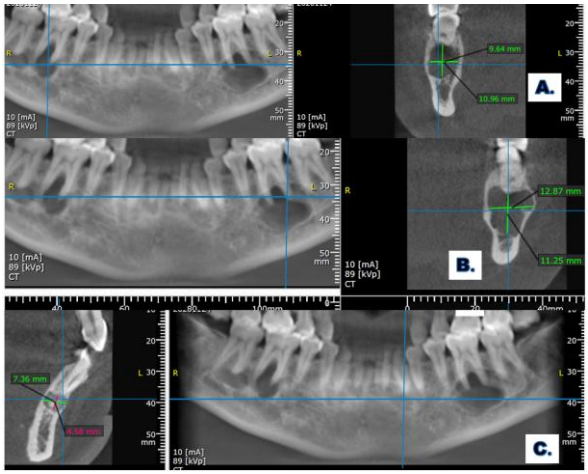

Fig. 2A. Axial CT revealed moderate bony expansion in posterior region of mandible along with thinning of buccal cortical plate irt 46 region. 2B: Axial CT revealed moderate bony expansion in posterior region of mandible irt 36 region. 2C: Axial CT revealed mild irregular bony expansion in anterior region of mandible

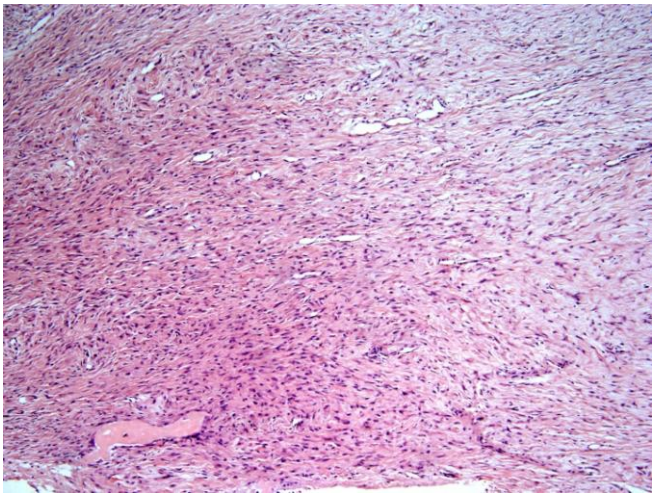

Fig. 3. Fibrocellular connective tissue with numerous angular, oval and stellate shaped cells, numerous fibroblasts, irregularly arranged collagen fibers and numerous round to oval cementum like spherules (10X)

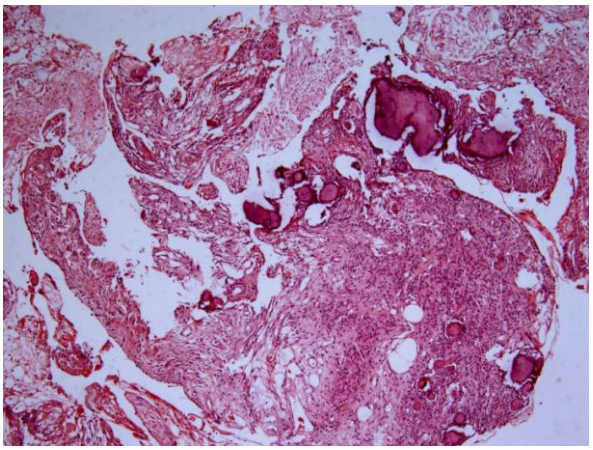

Fig. 4. Numerous round to oval cementum-like spherules in fibrocellular stroma (40X)

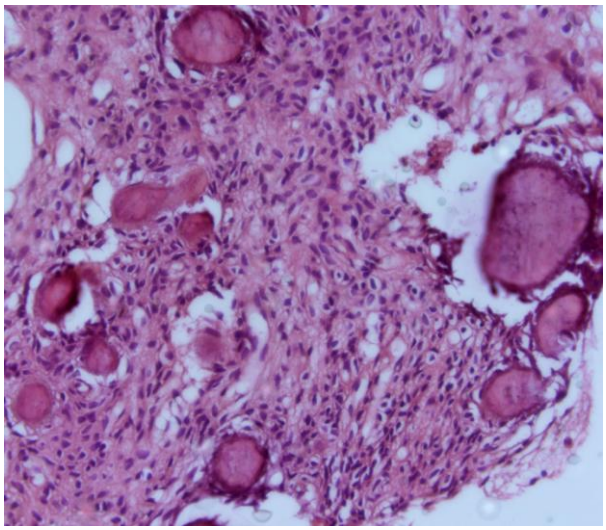

Fig. 5. Fibrocellular connective tissue with numerous fibroblast, irregularly arranged collagen fibres and eosinophilic osteoid like area (10X)

Present findings resemble a localized osseous reaction to chronic infection of the periapical tissue. “Pulpal inflammation may also stimulate epithelial rests in the periodontal ligament that experience subsequent liquefactive necrosis to form periapical or radicular cysts. Both of the above reveal spherical or ovoid radiolucency at the root apices of teeth, with variable peripheral radiopaque rim” [3]. In contrast, periapical abscesses and cysts almost invariably involve non vital teeth whereas in this case vitality test confirmed the vitality of all the lower teeth.

“Diffuse sclerosing osteomyelitis (DSO) is a primary inflammatory condition of chronic intraosseous infection with no odontogenic source and with soft-tissue swelling, fever and lymphadenopathy” [4-7]. It represents as wide, delineated sclerosis with osseous expansion and periosteal reaction. Unlike FCOD, unilateral mandibular involvement and pathologies are not confined to tooth-bearing areas in DSO. The basal and alveolar process of the mandible involved in DCO whereas FCOD was largely confined [6] that was evident in this case as both anterior as well as posterior regions were involved.

“Fibrous dysplasia (FD) is a localized, non-neoplastic characterized by osteoblast differentiation and maturation that may affect the multiple bones including jaw, frontal, ethmoidal, temporal, and calvarial bones. Most of the cases are monostotic and unilateral. FCOD characteristically exhibits bilateral bony expansion. FD more commonly affects the maxilla than the mandible and frequently involves adjacent bones such as the zygoma and

sphenoid and superior displacement of the mandibular canal is considered pathognomonic which was not evident in this case whereas FCOD commonly effects mandible and inferior displacement of mandible is seen in some cases" [3,7].

3. DISCUSSION

"Fibro-osseous lesions are a diverse group of entities in which normal bone is replaced by fibrous connective tissue and osseous components. Involvement of the jaw is seen in fibrous dysplasia, ossifying fibroma, and cemento-osseous dysplasias. The cemento-osseous dysplasias (CODs) represent non-neoplastic fibro-osseous lesions that arise exclusively in tooth-bearing areas of the jaw, arise from epithelial rests of the periodontal ligament" [3,8]. "Based on the location and extent of jaw involvement CODs divided into three categories: focal, periapical, and florid cemento-osseous dysplasia. If pathologies with similar radiologic and microscopic features involve two or more quadrants of the jaw, the disease is termed florid cemento-osseous dysplasia (FCOD)" [3,9].

"FCOD is a reactive fibro-osseous lesion confined to the alveolar areas of the jaws a typical female gender predilection affecting black women in 4-5 decades with a mean age of 42 years. These lesions were also seen in oriental populations and Caucasian females" [3-5].

FCOD can affect any tooth-bearing area, although it is proportionally more common in the mandible which is consistent with our findings. In a comprehensive analysis published in 2003, MacDonald-Jankowski found that 100% of instances involve the mandible, with a lower percentage (61.4%) affecting the maxilla. The posterior regions, particularly the mandible, are higher. The analysis of divergence of the maxilla and the front mandible had high rates, which may be explained due to the challenges in assessing these areas due to the superimposition of the anatomical structures that are above the surface [10].

FCOD probably occurs due to disorder in the differentiation of periodontal ligament stem cells resulting in irregular cementum formation [5]. "Some authors recognise the proliferation of the fibroblastic mesenchymal stem cells in the apical periodontal ligament which are cementoblastic precursor stem cells, while others accredit that it may arise from the remnants of the cementum remaining after tooth extraction" [11]. Waldron [12] proposed that "reactive or dysplastic changes in PDL might be the cause". "Some authors propose that trauma from deep bite or heavy bite causing attrition of the teeth may activate and cause proliferation of the fibroblasts in PDL causing FCOD" [11].

Pathologically, bone tissue is resorbed and replaced with fibrosed connective tissue composing fusiform and polyhedral cells. These irregular areas represent osteolytic radiolucent areas. At later stages these polyhedral cells which resemble undifferentiated mesenchymal cells initiate the deposition of disorganized collagen matrix, over which basophilic mineralized bone or cementoid begins to form. These areas represent radiolucency with irregular radiopacities that gradually unite to form radiopaque masses [6].

"Radiographic image of FCOD exhibits lobular radiopacities surrounded with radiolucent area and located mostly in the mandibular premolar-molar region. As the lesion matures it presents different radiographic features. The classic appearance reveals diffuse, lobular, irregular shaped radiopacities surrounded by ill defined radiolucency throughout the alveolar process, occurring above the mandibular canal" [1,13]. In the present case, defined radiolucency surrounded by thin radiopaque rim along with conglomerate radio-opaque masses were noticed on the left side of the mandible. Mixed radiolucent and radio opacity with ill defined borders was seen on the right side of the mandible and ill defined radiolucency was also evident in anterior region of the mandible. This implies various stages of maturation in a lesion and large confinement of the lesion.

FCOD can present radiographically as a completely radiolucent lesion in the early stages, before transitioning to a mixed radiolucent and radiopaque appearance in the intermediate stage, and finally becoming completely radiopaque, usually with a radiolucent rim, in the later stage of the illness. This aspect could be connected to the difficulties in identifying radiolucent lesions as a characteristic FCOD or to the fact that the diagnosis is made when patients seek conventional dental treatment and require a panoramic radiography for planning. Furthermore, symptoms are more common in the intermediate and advanced stages, which may lead to the patient seeking therapy and being diagnosed at these stages [14].

There is no need for any treatment in an asymptomatic patient, but regular follow up is mandatory due to the susceptibility to infection and fracture of the jaws, re-evaluation with panoramic radiographs should be done every 2 or 3 years in an asymptomatic patient. In symptomatic cases, administration of antibiotics is indicated to overcome secondary infection but sometimes it may not respond to antibiotics due to poor tissue diffusion [3-5,1] requiring surgical debridement and enucleation [1]. Therefore, management is focused toward prevention of exposure of avascular bone which causes subsequent development of osteomyelitis. For this reason, surgical procedures should be avoided where possible [15,16]. However, the need for surgical intervention to attain improved aesthetics and function should be evaluated depending on the case.

4. CONCLUSION

Florid osseous dysplasia is a benign lesion of tooth-bearing regions. This is mostly asymptomatic and coincidentally identified in routine dental imaging such as ortho pantograms. Differentiation from radiographically similar entities can avoid unnecessary and potentially harmful interventions or biopsy. Uncomplicated FCOD may remain asymptomatic FCOD for an indefinite period, doesn't have any complications and requires no invasive therapy but optimum oral hygiene has to be maintained to avoid tooth loss and periodontal disease. When the patient is symptomatic secondary to a tooth pain, the tooth may be managed endodontically by avoiding extractions. However, these hypo vascular, sclerotic lesions are prone to secondary infection when exposed to oral pathogens, which can be difficult to manage. Hence elective intraoral procedures have to be avoided.

CONSENT

As per international standard or university standard, patient(s) written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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