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Case Report

A Case of Florid Cemento-Osseous Dysplasia, Encompassing Both Maxilla and Mandible

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Abstract

Background: Florid cemento-osseous dysplasia (FLCOD) is a benign fibro-osseous lesion which changes cancellous bone tissue with each fibrous tissue and cementum-like material, or both together. The lesion most commonly affects the lower jaw involvement and rarely involving the maxilla. Lesion is usually asymptomatic and as a result an incidental finding during a routine dental radiographic examination. Once diagnosed, the lesion must be differentiated with other overlapping diseases to avoid the misdiagnosis followed by wrong treatment.

Case presentation: this paper aims to report a case describing the radiological features and proving the diagnostic workup of patients with florid osseous dysplasia (FLCOD). In current case a 42 year old lady presented with periodontal problem and routine radiograph investigation revealed multiple radio opacities in relation to periapical region of several teeth. This clearly emphasizes the importance of investigation, without which the case would be misdiagnosed. We therefore propose a routine radiological investigation for any systemic or generalized oral diseases to assist the clinician in the diagnosis of lesions.

Conclusion: Initial and later stages of FLCOD, overlaps with periapical diseases and others like paget's disease, chronic diffuse osteomyelitis, and Gardner's syndrome. Diagnosis and management of such confusing presentations must hence follow a methodology to avoid needless iatrogenic dental treatments and possible disease-related complications.

Keywords: Florid Osseous Dysplasia; Florid Cemento-Osseous Dysplasia; Homogenously Radiopaque; Cementum-Like Material

Abbreviations

(CODs): Cemento-Osseous Dysplasias; (FCOD): Focal Cemento-Osseous Dysplasia; (PCOD): Periapical Cemento-Osseous Dysplasia; (FLCOD): Florid Cemento-Osseous Dysplasia

Introduction

Cemento-osseous dysplasias (CODs) are fibro-osseous lesions where the normal bone architecture is replaced by fibroblasts and collagen fibers containing variable amounts of mineralized material [1].

World Health Organization (WHO) classification of cementomatous lesions published in 2005 includes cemento-ossifying fibroma, benign cementoblastoma and cemento-osseous dysplasia (COD) groups. Within COD lesions, 3 distinct subgroups of lesions exist, based on location and number of lesions. These 3 subgroups consist of focal cemento-osseous dysplasia (FCOD), periapical cemento-osseous dysplasia (PCOD), and florid cemento-osseous dysplasia (FLCOD). FLCOD was first described by Melrose., *et al*. in 1976 [2-4].

CODS are commonly asymptomatic and located in the periapical region of teeth with pulp vitality and are usually noted in routine radiologic exams. The posterior tooth-bearing regions are the usual sites of involvement; the maxilla is rarely affected. Severe cases with infection presents, dull pain, drainage, exposure of the lesion in oral cavity, focal expansion and facial deformities [5,6]. Whereas it's more frequent in females as compared to males, it is generally seen within the fourth and fifth decades. Radiographically, FLCOD is characterized by multiple masses of mixed radiopaque structures. Often with a circumferential radiolucency, primarily surrounding the root apices of vital teeth, and over time with the maturation of the lesions, radiographic images can become increasingly radiopaque [7].

The diagnosis of COD is usually made by correlation of demographic information (age, gender and ethnicity), clinical (location and number of lesions) and investigatory features (image type and histopathology) [8]. The diagnosis based on these features is necessary because, clinically the pathology share similar characteristics with that of other lesions. This might lead to misdiagnosis, at various stages of development [9,10]. Hence a thorough clinical knowledge and investigations is essential for accurate diagnosis, which will further aid in proper management of disease. In this paper, a case of a 45 year female patient presenting with signs of periodontal disease, was later diagnosed with FLCOD on the basis of clinical, radiographic and biochemical findings.

Case report

A 45 year old female patient reported with a complaint of missing upper left front tooth and wants replacement of the same. She also complains of mobility of upper right front tooth since 3 months. She gives history of exfoliation of teeth 1 year back following mobility. Mobility was insidious in onset and gradually progressive. She is known diabetic since 6 months and on medication every day. Her extra oral examination revealed bilateral solitary submandiblar lymph nodes palpable and enlarged, mobile and tender. On intra-oral examination there was presence of generalized gingival inflammation, generalized gingival recession and periodontal pockets (Figure 1). Hard Tissue Examination shows clinically missing teeth 12,18,21,26,28,31,32,37,38,41,42,47,48, wear facet with tooth 13, 23 and mobility Grade I in relation to teeth 14, 24, 25, 27, 36, 46, Grade II mobility with tooth 15, 17 and Grade III mobility with tooth [11].

Figure 1: Intra oral examination reveals generalized gingival recession and multiple missing teeth.

Investigations

Since the patient had extensive periodontal problem she was subjected to routine Orthopantomograph. Which showed multiple radiopacities in the periapical region and thus the patient was further subjected to Vitality Testing of involved teeth, Random Blood Sugar, Serum Biochemistry to rule out other bony diseases and finally a portion of lesion was sent for histopathology to confirm the diagnosis.

Radiographic description of the lesion

Location

The focal radio opacities are present on the alveolar bone in relation to the periapical region of the teeth as well as non tooth bearing region of the maxilla as well as mandible, both anteriorly and posteriorly. All the radio opacities are located above the inferior alveolar canal in the mandible.

Investigations	Result	Reference range
Orthopantomograph (Figure 1)	**Reveals,** Generalized horizontal alveolar bone loss of about >4-5 mm below the Cemento-enamel junction. Angular bone loss with tooth 15, 36. And multiple radio opacities in relation to periapical region of multiple teeth.	
Vitality Testing (Figure 2,3 and 4)	All the teeth were tested for vitality using heat test. They were found to respond normally suggestive of vital teeth.	
Random Blood Sugar	75 mg%	70 - 110 mg%
Serum Biochemistry		
• Acid Phosphatase	4.9 IU/L	0-6 IU/L
• Alkaline Phosphatase	221.2 IU/L	80-306 IU/L
• Calcium	7.9 mg%	8.4 - 10.2 mg%
• Inorganic Phosphorous	3.9 mg%	2.6 - 4.5 mg%

Table 1: Diagnostic workup in FLCOD.

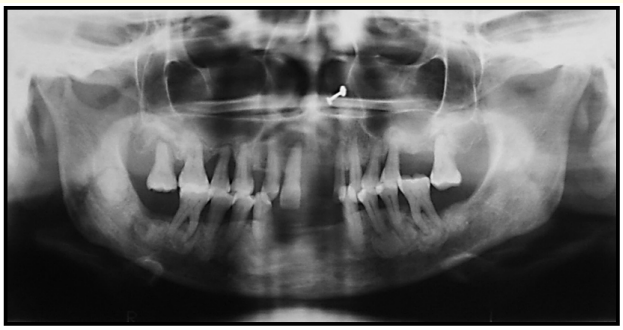

Figure 2: OPG reveals generalized horizontal alveolar bone loss and multiple radio opacities in relation to periapical region of multiple teeth.

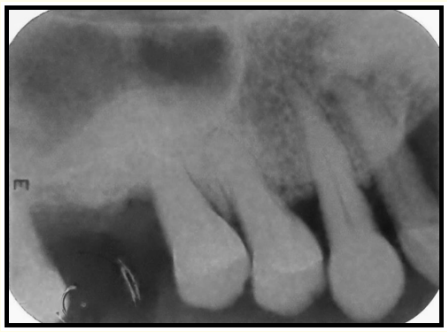

Figure 3: IOPA 24, 25 displays ill-defined radiopacity blending with the surrounding bone.

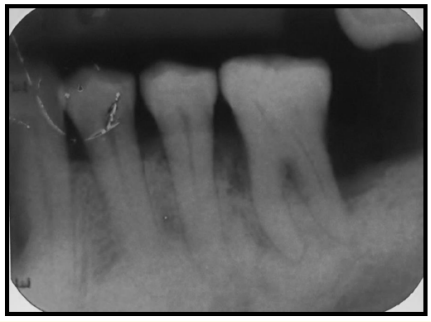

Figure 4: IOPA with 34, 35 and 36 demonstrated a radiolucent lining around the radiopaque mass suggestive of a soft-tissue capsule

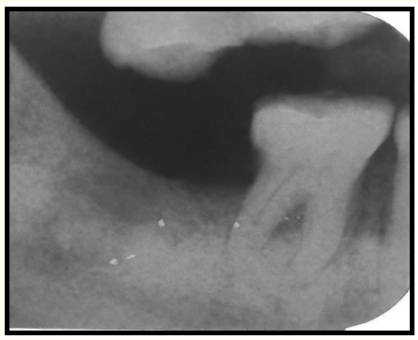

Figure 5: IOPA with 46 presents with well-defined radiopacity without distinct radiolucent lining

Internal structure

The focal lesions in the maxilla seemed to have homogenously radiopaque internal structure. The mandibular lesions on the other hand seemed to have interspersed radiolucent areas amidst the radiopacities.

Periphery

The peripheries of the focal lesions vary from one region to another. In relation to 14 as well as 16 they appear distinct but without the evidence on a radiolucent lining. In relation to 24, 25 (Figure 2) they appear ill defined and the radiopacity seems to blend with the surrounding bone. However in relation to the mandibular lesions, they demonstrated a radiolucent lining suggestive of a soft-tissue capsule especially in relation to 34, 35 and 36 (Figure 3). On the right mandible (Figure 4), the periphery was well defined but a distinct radiolucent lining could not be appreciated. A solitary focal radiopacity was noted on the right ramus region which demonstrated a well-defined radiolucent periphery (Figure 1).

Effect on surrounding structures

The lesions seemed to have no effect on the surrounding structures. Neither the maxillary sinus, nor the mandibular canal showed any displacement. There was no resorption or displacement of the roots of the corresponding teeth.

Histopathology report

A solitary lesion was excised at the edentulous area in relation to the missing tooth 48 for histopathologic examination. Histologic findings reveal the formation of calcified dense sclerotic cementum-like masses. Anastomosing bone trabeculae and layers of cementum-like calcifications are embedded in a fibroblastic background.

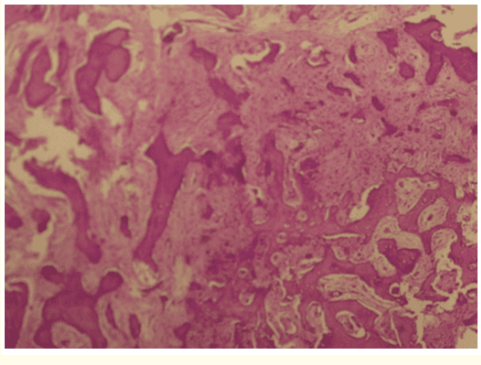

Figure 6: showing anastomosing bone trabeculae and layers of cementum-like calcifications embedded in a fibroblastic background

Final diagnosis and differentials

On the basis of the patient's clinical, radiographic and histopathologiocal findings a final diagnosis of Florid Cemento-osseous dysplasia was given with differential diagnosis of multiple focal cemento- osseous dysplasia, Paget's disease, chronic diffuse osteomyelitis, and Gardner's syndrome.

Treatment

We decided to curb the surgical intervention, since the lesion was multifocal and diffused. Patient was kept under regular clinico-radiological follow-up. Besides that, grade II, III mobile teeth 17, 15, 11 were advised for extraction followed by oral prophylaxis and replacement of missing teeth with removable partial denture.

Prognosis

Patients with florid cemento-osseous dysplasia do not generally have any symptoms. They may get infections in the teeth and jaw, which can be treated by surgery by removal of the bony lesion if it's localized. The long-term outlook with this condition is generally good.

Discussion

The current classification of bone-related lesions, released in 2005 by the WHO, is based on age; sex; histopathological, radiographical and clinical characteristics along with location of the lesion [11]. Eversole., *et al*. described the various fibro-osseous lesions identified within the craniofacial complex.

The lesions are thought to arise from inflammatory, reactive, neoplastic, or developmental dysplastic disease processes, whereby normal osseous tissue is replaced by fibrous tissue interspersed with ossifications and calcifications in a progressive manner [12,13].The lesion is related to the teeth, even though it is not considered to be odontogenic. Nearly all FLCODs occur above the mandibular canal and are thus confined to the alveolar process, suggesting at least some odontogenic influence on their genesis [1,14]. This remains true in current case, where the lesions were above mandibular canal and close to maxillary sinus, without affecting the vital tissues but related to apical region of the teeth.

The molar and premolar teeth are the most frequently involved. Lower jaw involvement is around 94% and sometimes affecting the maxilla. FLCOD presents with cortical expansion, particularly of the mandible. And is usually asymptomatic and as a result an incidental finding during a routine dental radiographic examination [15]. Radiographs show large, radiolucent, mixed, or most often, dense radiopaque masses, limited to the periapical alveolar bone. They do not involve the inferior border, except through direct focal extension, and do not occur in the rami [16].

The rare clinical symptoms observed are pain, swelling and local drainage but these are only encountered in cases of secondary infection, when the calcified masses are exposed in the oral cavity. Women are more affected than men and the disease occurring mostly around the fourth decade. The lesion follows an ethnic distribution, 59% in African patients, 37% in Asians and 3% in Caucasians [17]. Similarly in the present case, the lesion is seen in 45 year old Asian female, with complain of mobile tooth giving a clinical picture of periodontal problem and FLCOD was an accidental finding on routine radiographic examination and was an extensive form of lesion, involving maxilla and mandible along with anterior and posterior teeth.

The early stage of FLCOD presents with a well-defined radiolucency at the apices of teeth. The radiographic image may be misdiagnosed as an endodontic infection, without leading to either tooth displacement or root resorption. A pulp vitality test should be performed to rule out any endodontic infection [18]. Our case presented the later stage of FLCOD with diffuse radiopacity, often with ill-defined borders and a greater proportion of anastomosing, thick and poorly cellular bony trabeculae. FLCOD should be differentiated from Paget's disease, chronic diffuse osteomyelitis, and Gardner's syndrome to avoid misdiagnosis. Jong-Ki Huh., [19] *et al*. presented a similar case of FLCOD with unnecessary endodontic treatment due to misdiagnosis. The difference is that FLCOD is seen above the inferior alveolar canal, whereas Paget's involves the entire mandible along with loss of lamina dura. Moreover, biochemical serum changes such as elevated alkaline phosphatase levels occur in Paget's. In our case, radiographs showed that the inferior alveolar canal and maxillary sinus weren't affected, and there was no serum alkaline phosphatase level change. Gardner's syndrome also presents with other skeletal changes, skin tumors and dental anomalies, but FLCOD is only seen at tooth-bearing areas. Differentiation from chronic diffuse osteomyelitis is also similar because osteomyelitis shows single area of diffuse sclerosis containing small, ill-defined osteolytic areas and involves the body of the mandible from the alveolus to the inferior border and may extend into the ramus. While FLCOD, presents with multiple round or lobulated radiopaque masses, involving the apices of teeth. Fibrous dysplasia has pathognomonic ground glass appearance in radiologic images; additionally, FLCOD is not a developmental lesion unlike fibrous dysplasia [20-22].

As FLCOD is a benign usually self-limiting disease, management of such condition mostly entails simple monitoring of the lesions, with an annual long-term clinical and radiographic follow-up [23]. Same was monitored in our case, avoiding surgery, due to the multifocal and diffused involvement of the lesion. A solitary lesion was excised in the current case at the edentulous area in relation to the missing tooth 48 for histopathologic examination. The histopathological appearance of FLCOD consists of bone trabeculae and cementum-like material present within a vascular fibrous stroma [24], that are consistent with those of our case, thus confirming the diagnosis of FLCOD.

Conclusion

Although FLCOD is generally asymptomatic and non-neoplastic, sometimes it can reach large sizes. It may cause expansion and/or perforation of the alveolar bone. Early and later stage of FLCOD overlap with many other diseases as discussed above and presents dilemma to clinicians regarding its exact nature and pathogenesis. Therefore it is important to be familiar with its radiographic and clinical presentation. Where, the role of the oral medicine and maxillofacial radiologist in the differential diagnosis is an essential one. Diagnosis and management of such confusing presentations must hence follow a methodology to avoid needless iatrogenic dental treatments and possible disease-related complications.

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