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Review Article

Florid Cemento-Osseous Dysplasia – A Rare Case with Diverse Radiographic Presentation

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Abstract: Florid cemento osseous dysplasia (FCOD) is a rare, benign lesion affecting both the jaws. It is found more commonly in middle - aged black females and also in Caucasians and Asians. It is characterized by substitution of bone with cement-osseous tissue. Generally asymptomatic, FCOD is typically an incidental finding in routine radiographs. Radiographic features depend on the developmental stage of the lesion which may be radiolucent, mixed or radio opaque. Usually presents bilaterally symmetrical in association with the periapical area of the dentition. May present with multiple impacted teeth in the jaw and has a slight familial distribution. Hereby we present a case of FCOD and multiple impacted teeth with a radiographic presentation of all the three stages of formation.
Keywords: Florid, rare, pathologic fracture, asymptomatic, conservative, multiple impacted teeth, Gigantiform Cemetoma

INTRODUCTION

Cemento osseous dysplasias (COD) are a group of rare, benign, generally asymptomatic dysplasias of the jaw bones originating from periodontal tissue. World Health Organization classified COD based on age, sex, histopathological features, radiological characteristics and location of the lesion. This classification includes the likes of cemento-ossifying fibroma, benign cemento blastoma and cemento-osseous dysplasia (COD) groups. COD are further classified as Periapical (Peripaical region of tooth and generally bilateral), Florid (sclerotic symmetrical masses) and Focal (Single lesion) [1].

Florid Cemento Osseous Dysplasia is a subdivision of COD. FCOD has been called along the years with various names like Gigantiform Cemetoma, Multiple Enostosis, Multiple Cemento-Ossifying Fibroma, Sclerosing Osteitis, and Sclerotic Cemental Masses of the Jaw [2]. FCOD was first described by Melrose *et al.;* in 1976 [3]. Florid according to Oxford dictionary means something "Excessively intricate or elaborate". Thus FCOD are extensive sclerotic lesion found in multiple quadrants of both the jaws of a dentate or edentulous patient. It is mainly found in middle-aged black female populace (about 90 %) although some predilection is seen in Caucasians, Asians [2] but only about 2% of cases are seen amongst Indians [4]. Familial tendency are generally not seen except in few secluded cases [5]. Etiology is mostly unknown although a periodontal origin is widely accepted [6].

Diagnosis of FCOD is primarily a radiographic one although clinical features should be correlated along with a steady follow-up. The lesions are typically asymptomatic and are incidental radiographic findings. Symptoms may appear as dull aching pain or mild expansion of cortex or pus discharge if the sclerotic masses get exposed to the oral atmosphere. Hence an ill-fitting dentures, biopsies and elective extraction are contraindicated as they may prompt an infection or lead to jaw fractures [8]. Thus treating a FCOD aggressively might lead to mediocre outcome causing dissatisfaction to the patient. A preventive approach with regular follow ups, maintenance of good oral hygiene and periodontal status should be followed [2].

CASE REPORT

A 42 year old, moderately built, well-nourished Indian female patient reported to the Department of Oral and Maxillofacial Surgery with chief complaint of multiple missing teeth and root stumps. Patient had no significant medical or family

history. Upon clinical examination of the oral cavity multiple root stumps were noted. 47, 48 and 38 were found to be missing with no history of any previous extraction. No obvious swelling was found. Orthopantomographic examination (Figure) revealed multiple impacted teeth (47, 48, and 38). Well defined, irregular sclerotic masses were noted below 48, 47, 18 region whereas a mixed appearance (area of radiolucency surrounded by a rim of radioopacity) was seen in the 44, 47 region. A periapical radiolucent mass was seen near 45 region. The impacted 47 along with its periapical sclerotic mass involved the lower border of mandible. There was no evidence of root resorption or any carious lesion in the involved teeth. Lamina dura was seen intact. All the clinical and radiological evidence resulted in the diagnosis of a case of Florid Cemento-osseous dysplasia. The root stumps were extracted. The dysplastic masses and the involved teeth were not removed as they were symptomless. Patient was kept on regular follow- up.

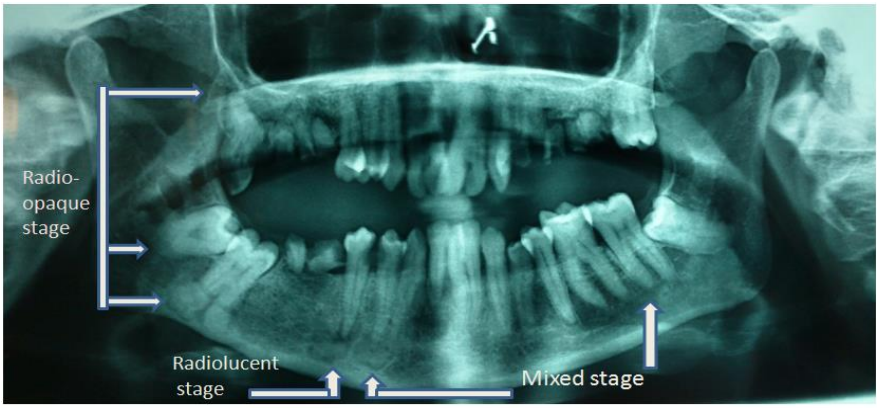

Fig-1: OPG depicting all three stages of FCOD associated with different teeth and multiple impacted teeth

DISCUSSION

FCOD is a rare disease causing replacement of bone with dysplastic, cementum like substance in the jaw. It affects more than one quadrant of the mouth. Melrose first described the lesion in 1976. Waldron proposed the term Florid Cemento-osseous dysplasia. Robinson gave it a definition saying that it is an abnormal reaction of bone to irritation or other stimulus. Melrose highlighted the non-inflammatory character of the lesion as it fails to resolve even after removal of irritants. Eversole and Cho *et al.;* was in favour of the progenitor role of periodontal ligament for the adjacent hard tissue. Few authors have been able to link a familial tendency to the condition, with an autosomal dominant trait with a variable phenotype. Certain literature reveals a co-occurrence of multiple impacted teeth with FCOD and most of them having a familial tendency [9, 5].

FCOD is a rare condition with an unknown etiology. Mostly found in Black – middle aged females, though also found in Caucasians and Asians. 2 % cases are found in Indians [2, 4]. Clinically it is asymptomatic, self-limiting, non-expanding lesion. It affects both jaws, crossing the midline and are bilaterally symmetrical. They are found only on the dentate individuals, hinting to its reactive origin. It is generally a chance finding in scout radiographs. Some non-specific symptoms like dull gnawing pain, purulent discharge – due to exposure of the sclerotic masses into the oral cavity, may be found. In rare cases an expansile mass that dilates the cortical plates, causing facial deformity, are seen [10].

FCOD is thought to be originating from the periodontal ligament tissue, when the normal vascularised bony tissues are transformed to avascular cementum like lesion. Pathology is ambiguous. Some school of thought preaches the proliferation of fibroblastic mesenchymal stem cells in the apical periodontal space, which are precursor of cementoblasts, to be responsible [9]. Others view it as vestigial remnants of cemental tissues following tooth extraction. Certain predisposing factors like trauma, carious lesion, periodontal disease, trauma from occlusion, deep bite, systemic illness, hormonal imbalance may play a major role in the onset of the condition [1, 6].

Table-1: Radiographically these lesions can be classified into three distinct stages, depending on their development [7]

STAGE	RADIOLOGICAL FEATURE
Early / Osteolytic stage	Well defined radiolucency with loss of periodontal ligament and lamina dura
Intermediate / cemento blastic stage	Small radiolucent centre with a radioopaque rim, often misdiagnosed as cement-ossifying fibroma.
Late / Osteosclerotic stage	A definite radioopaque, sclerotic mass observable.

Histopathologically, all the stages will show similar features. It is a heterogenous lesion consisting of a benign fibrous stroma with irregular trabeculae of mature and immature bone and cementum like tissue. As the lesion matures there is an increase in mineralization. Trabeculae become arciform and cemental masses fuse to form large basophilic structures [9].

Various similar disorders may mimic FCOD cause a diagnostic doldrum for the doctor. A periapical granuloma or cyst may be look like the osteolytic stage of FCOD but are generally associated with a carious tooth or after a tooth is injured due to trauma. Chronic Diffused Sclerosing Osteomyelitis, Cemento Ossifying Fibroma, Odontoma, Osteoblastoma may resemble the Intermediate stage of FCOD. Certain systemic diseases like Gardner Syndrome, Paget's disease falls in a similar spectrum of symptoms. In Gardner syndrome one can see skeletal changes, skin tumours unlike FCOD. An increase in serum alkaline phosphatise is diagnostic of Paget's disease not evident in FCOD. A bilateral expansion is more common in Cemento Ossifying Fibroma than FCOD. In Chronic Diffused Sclerosing Osteomyelitis systemic illness like fever, lymph adenopathy are seen and are generally unilateral, affecting mostly the mandible. Excess bony expansion comprising of a radiolucent rim is seen more in osteoblastoma than FCOD [10].

If the patient is asymptomatic then no treatment is required. Regular follow-up visits are recommended to ensure that the lesion is within normal limits. In case of any symptoms like purulent discharge and sinus tract formation, surgical resection and saucericzation is advised. Complete resection is generally contraindicated as the lesion involves the complete jaw and may increase morbidity. Biopsies are generally disdained as they may increase chances of infection and pathologic fracture. Extractions are avoided in view of poor socket healing due to diminished blood supply [10].

CONCLUSION

FCOD is a rare condition that a dental surgeon comes across. It is a rare incidental finding in routine radiographs. Treatment of such disease varies from patient to patient. Conservative management should be the first line of treatment due to the asymptomatic nature of the lesion. Aggressive surgical management may cause more harm than good. Thus one must carefully weigh the benefit to risk ratio while treating such lesions.

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