Florid Cemento-Osseous Dysplasia with Multiple Impacted Supernumerary Teeth in Maxilla and Mandible – A Case Report

Jayanta Chattopadhyay[1], Soumi Ghanta[2]

ABSTRACT

Introduction: Florid cement-osseous dysplasia is a benign, fibro-osseous and multifocal dysplastic lesion of the jaw and consists of cellular fibrous connective tissue with bone and cementum-like tissue.

Case report: This clinical report describes a nonfamilial case of Florid cemento osseous dysplasia (FCOD) accompanied by osteomyelitis in a 41-year old male patient. Orthopantomograph revealed multiple radiopaque cemetum-like masses distributed throughout the maxilla and mandible. There were multiple impacted supernumerary teeth present in both jaws. Blood investigation did not detect any abnormality in serum alkaline phosphatase level. Antibiotics were given for osteomyelitis. No impacted teeth were extracted as they were asymptomatic. After that, patient is followed up at regular intervals with reinforcement of good oral hygiene to prevent osteomyelitis.

Conclusion: Clinicians should be aware of the radiographic manifestation of such conditions and knowledge to recognize and differentiate from other conditions similar to their appearance.

Keywords: Florid-cemento osseous dysplasia, Fibroosseous lesion, Imapcted teeth, Osteomyelitis.

INTRODUCTION

The term osseous dysplasia refers to the abnormal development and disordered production of bone and cementum like tissue. Waldron considered these lesions to be fibroosseous lesions arising from periodontal ligament. These osseous dysplasias are categorized as periapical (surrounds the periapical region of tooth), focal (single lesion) and florid cemento osseous dysplasias. The word 'florid' refers to wide spread, extensive manifestation of the lesions. Florid cemento-osseous dysplasia (FCOD), previously called *gigantiform cementoma, multiple cemento-ossifying fibroma, sclerosing osteitis, multiple enostosis* and *sclerotic cemental masses of the jaws*, was first described by Melrose et al.[1] It is a benign, fibro-osseous and multifocal dysplastic lesion of the jaw and is usually asymptomatic and occurs most often bilaterally. It consists of cellular fibrous connective tissue with bone and cementum-like tissue.[2,3] It may/may not cause cortical expansion. There is a tendency for secondary infection leading to osteomyelitis due to avascular nature of these bony lesions.[2]

A search of the literature showed that only a few cases have been reported concerning the familial form of FCOD associated with multiple impacted teeth. However, very few examples were found of the nonfamilial form of FCOD associated with multiple impacted teeth. Here we present details of a very rare case of nonfamilial FCOD associated with multiple impacted teeth in both jaws accompanied by osteomyelitis.

CASE REPORT

A 41 year-old male patient reported to the dental department of Narayana Multispeciality Hospital with a complaint of a pain and discharging of pus in the left lower posterior teeth region. He gives history of extraction of the mandibular left posterior teeth two years back due to mobility and pus discharge. The extraction event was associated with delayed healing. Patient also gives previous history of extraction of maxillary left anterior and posterior teeth and mandibular anterior teeth 3 years back due to caries. Family and medical history were non-contributory. There was no history of swellings or bony deformity elsewhere in the body. The familial history was taken and some of the family members were examined, but no familial aspects of the disease could be established. On extraoral examination, facial asymmetry was present and the length from angle of mandible to midline of left side was more than that of right side. Intraoral examination revealed a mild diffuse swelling extending from 36 to 38 region causing expansion of buccal cortical plate and mildly lingual cortical plate. On palpation it was hard and tender. There was discharging of pus present distal to 36 region (Figure-1). 37 and 38 were missing. Left lower first molar was tender to percussion. On further examination 23, 26, 43 were missing (Figures-2,3). However, the alveolar mucosa with respect to the missing teeth was normal. The bucco-palatal width of 22 to 26 region was more than that of right side (Figure-2). Orthopantomograph revealed multiple sclerotic masses scattered in both the jaws (Figure-4). These sclerotic masses were present above the inferior alveolar canal in mandible. The areas of mixed radiolucent and radiopaque pattern involving both the jaws are also notified. Moth eaten type of radiopacity was present distal to left lower first molar. There were multiple impacted supernumerary teeth present in both maxilla and mandible. Most of the impacted teeth seemed to be pushed through the periphery of the jaws by the expansile lesions. Blood investigations did not detect any biochemical abnormalities. Serum alkaline phosphatase was normal. Based upon this clinical and radiographic finding, this condition was diagnosed as nonhereditary type of florid cement-osseous dysplasia with osteomyelitis and multiple impacted teeth. Biopsy was not done as the case can be diagnosed on the basis of the characteristic features seen on the radiographs. Antibiotics were given for osteomyelitis. No impacted teeth were extracted

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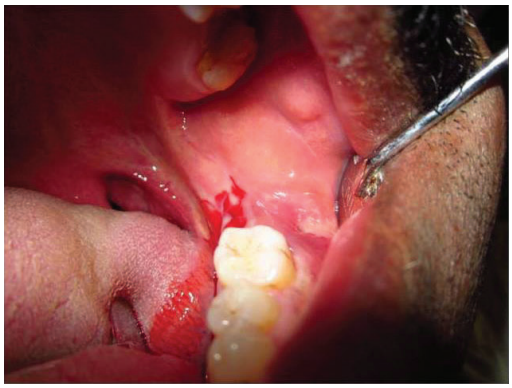

Figure-1: Intraoral view- Presence of pus discharge in left mandibular posterior region

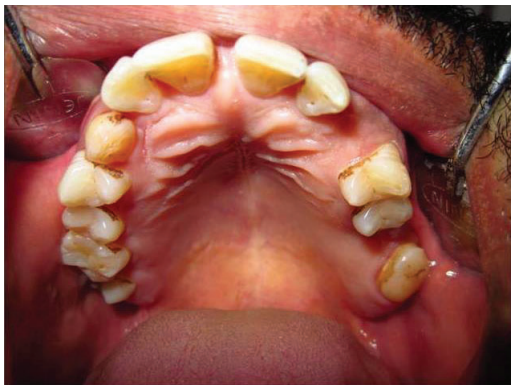

Figure-2: Intraoral view of maxillary arch- Missing 23 and 26; more bucco-palatal width from 22 to 26 region as compared to right side.

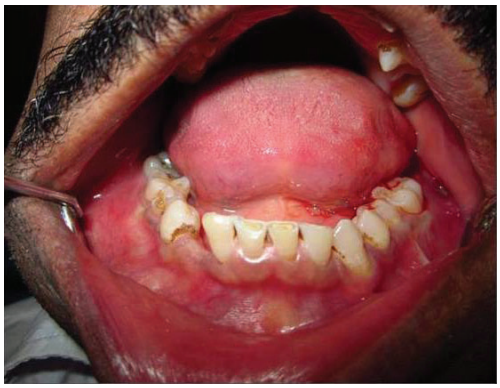

Figure-3: Intraoral view of mandibular arch

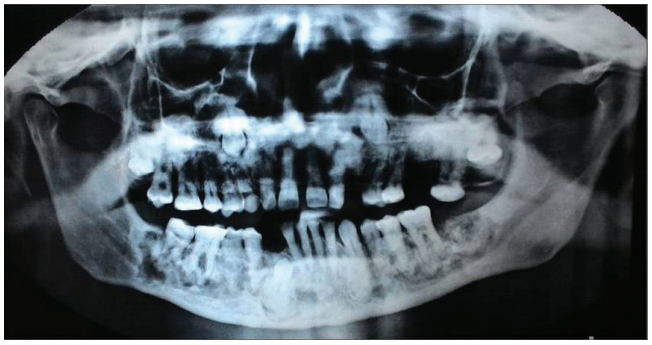

Figure-4: Orthopantomograph revealed multiple sclerotic masses scattered in both the jaws and moth eaten type of radiopacity was present distal to left lower first molar. Multiple impacted supernumerary teeth were also present in both the jaws.

as they were asymptomatic. Patient is followed up at regular intervals.

DISCUSSION

Exuberant fibro-osseous lesions occurring in multi-quadrants of the jaws were designated as gigantiform cementomas or familial multiple cementomas in the first edition of the World Health Organization's Histological Typing of Odontogenic Tumours, Jaw cysts and Allied Lesions.[4] Florid osseous dysplasia was first suggested by Melrose et al in 1976.[2,4,5] After that Waldron proposed the term florid cemento osseous dysplasia (FCOD) because of closely resemblance of dense sclerotic masses with cementum.[4] It is defined by Robinson to be an abnormal reaction of bone to irritation or stimulation. Melrose pointed out non-inflammatory feature of these lesions because they failed to resolve after elimination of presumed irritants. Eversole and Cho et al supported the progenitor role of periodontal ligament fibroblasts for adjacent hard tissue cells. These lesions are characterized by replacement of bone by connective tissue matrix in which the matrix displaying varying degrees of mineralization in the form of woven bone or cementum-like round basophilic acellular structures. FCOD is most commonly seen in middle aged females.[2,5,6] The present case of a 41-year-old male patient may represent the first of such a rare combination of features being reported in the Indian literature.

FCOD has a tendency for bilateral, symmetrical involvement. It occurs most often in the mandible, but may occur in all four quadrants of the jaws.[2,3,6] All four quadrants were involved in the present case. It is most often asymptomatic and detected through routine radiographic examination. Symptoms such as dull pain or drainage of pus may occur. This lesion is susceptible to osteomyelitis because of exposure of densely sclerotic cemental masses to the oral environment from progressive alveolar atrophy under a denture, traumatically induced ulceration of alveolar mucosa or tooth extraction or biopsy. So, biopsy is not indicated. Clinical and radiological features are most important for diagnosis. FCOD rarely shows cortical expansion[1-3,6] which was present in our case.

Few reported cases of familial form of FCOD have been found in the English literature which appears to be inherited as an autosomal dominant trait with variable phenotypic expression. Unlike the sporadic cases, the familial form is characterized by more expansile lesions and tends to occur in younger individuals.[5,7] In the present case, no familial aspects of the disease could be established. The case had multiple impacted teeth with cortical expansion in both jaws. The case was painless except left side of mandibular posterior region which was secondarily infected leading to osteomyelitis. Very few reported cases of FCOD with multiple impacted teeth were found and most of them were familial in nature.[2] The nonfamilial form of FCOD very rarely shows such a combination.

The radiographic appearance of FCOD depends on the degree of maturation of the lesion. The most common radiographic presentation is multiple confluent sclerotic masses admixed with well defined areas of a mixed radiolucent-radiopaque pattern located usually in the tooth-bearing regions. Over time, the lesions tend to become increasingly radiopaque. The lesion may present different radiographic features in different stages. The classic appearance includes diffuse, lobular, irregular-shaped radiopacities throughout the alveolar process.[1,2,5,8] They do not involve the inferior border, except through direct

focal extension and do not occur in the rami.[2,5] In the present case, multiple sclerotic masses were scattered in both the jaws. These sclerotic masses were present above the inferior alveolar canal in mandible. Computed tomography (CT) can be used to differentiate FCOD from lesions that exhibit a similar sclerotic appearance on conventional radiographs. Enostosis or exostosis exhibits well defined high-density masses on axial CT images than on occlusal radiographs, and they are found to be continuous with cortical plates.[5]

The histomorphology shows a spectrum of progressive features depending upon the stage of development. In the initial stages there is uncapsulated proliferation of cellular fibrous connective tissue containing numerous small calibre blood vessels. In the more advanced stages, mineralized tissue consisting of woven and lamellar bone and cementum like tissue may appear pagetoid with prominent resting and reversal lines.[2,6]

Biopsy is not indicated as FCOD lesions are susceptible to infection leading to osteomyelitis due to avascular nature of altered tissues. Clinical and radiological features are most important to diagnose FCOD.[2]

FCOD lesions should be differentiated from other similar appearing sclerotic lesions on conventional radiographs. Paget's disease manifests as cotton-wool appearance involving multiple bones such as maxilla, mandible, spine, femur, skull, pelvis and sternum and produces elevated alkaline phosphate levels whereas florid cemento-osseous dysplasia is centred above the inferior alveolar canal in mandible and serum alkaline phosphatase level is within normal limit. FCOD usually does not involve other bones. FCOD usually does not involve other bones.[2,5,7] No biochemical alterations and other bone involvement were found in this case presented. Differential diagnosis of FCOD should also include chronic diffuse sclerosing osteomyelitis, which can be a complication of the disease.[5] Chronic diffuse sclerosing osteomyelitis is a primary inflammatory condition of the mandible presenting with cyclic episodes of unilateral pain and swelling. It 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 is not confined to tooth-bearing areas. It may involve 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 arising in tooth-bearing areas.[2,5-7] This lesion does not appear to be developmental in nature such as fibrous dysplasia, nor does it show the characteristics of neoplasia such as ossifying fibroma.[3] Odontogenic tumors, especially cemento-ossifying fibroma, usually exhibit more buccolingual expansion than does FCOD.[4] Rare possibilities include Gardner's syndrome. Clinical examination and patient data may be crucial in differentiating the lesions. Gardner's syndrome, which can include multiple enostoses, usually is associated with intestinal polyposis and is hereditary. Multiple osteomas frequently are associated with them.[2,9] Florid cemento-osseous dysplasia has no other skeletal changes or tumours that are seen in this syndrome.[6]

There is no need for any treatment in an asymptomatic patient. Antibiotics are generally not effective in FCOD due to poor tissue diffusion, but regular follow up is mandatory due to the susceptibility to infection and fracture of the jaws.[2,5,8] Instructions to keep good oral hygiene should be given to control periodontal disease and prevent tooth loss. Reevaluation with panoramic radiographs should be done in every 2 or 3 years in an asymptomatic patient and dental CT imaging should be considered if new symptoms or signs develop.[3,5,6,9] No treatment is required unless the patient is esthetically concerned or becomes symptomatic. Management of the symptomatic patient is more difficult because of development of chronic inflammation and infection within densely mineralized tissue.[2,5,6] In such 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.[1,2,5] However saucerization and extensive surgical resection are probable treatment options for extensive and symptomatic lesions. All efforts should be made to preserve the teeth because of protracted healing after tooth extractions.[2,5] There is no satisfactory treatment for this condition till date.

CONCLUSION

Mostly florid cemento osseous dysplasias are asymptomatic and are found during routine radiographic examination. Clinicians should be aware of the radiographic manifestation of such conditions and knowledge to recognize and differentiate from other conditions similar to their appearance.

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