

Case Report

Florid Cemento-Osseous Dysplasia with Chronic Osteomyelitis: A Case Report

Ravinder Singh¹, Deepak Gupta¹, Amit Aggarwal¹, Soheyl Sheikh¹, Shambulingappa P¹, Neha Bansal¹, Jyoti Mago²

Oral Medicine and Radiology, ¹MM College of Dental Sciences and Research, Mullana, Ambala., ²SKSS Dental College & Hospital, Ludhiana.

Corresponding Author

Dr. Jyoti Mago
Senior Lecturer
Oral Medicine and Radiology
²SKSS Dental College & Hospital,
Ludhiana.
Email - jyotimago22@gmail.com

Received: 04-07-2014

Revised: 12-07-2014

Accepted: 16-07-2014

Abstract

Florid cemento-osseous dysplasia refers to a group I of fibro-osseous lesions which are exuberant, multi-quadrant and arise from the tooth-bearing area of the jaws that consists of cellular fibrous connective tissue with bone and cementum like tissue. The lesion is usually benign and requires no treatment unless cosmetically concerning or symptomatic. Symptoms such as dull pain or drainage are almost always associated with exposure of the sclerotic calcified masses in the oral cavity. This may emerge as the result of progressive alveolar atrophy under a denture or after extraction of teeth in the affected area involved. In this case report a rare case has been reported that occurred in middle aged Indian women of a secondary osteomyelitis of a Florid Cemento Osseous Dysplasia lesion during the surgical procedures and the diagnostic challenges it posed along with its treatment options.

Key Words: Fibro-Osseous Lesions; Florid Cemento Osseous Dysplasia.

This article may be cited as: Singh R, Gupta D, Aggarwal A, Sheikh S, P Shambulingappa, Bansal N, Mago J. Florid Cemento-Osseous Dysplasia with Chronic Osteomyelitis: A Case Report. J Adv Med Dent Scie Res 2014;2(3):146-150.

Introduction

The term florid cement-osseous osseous dysplasia (FCOD) was first suggested by Melrose *et al* in 1976 to describe a condition of exuberant multi quadrant masses of cementum and/or bone in both jaws and in some cases, simple bone cavity like lesions in affected quadrant.¹ The word 'florid' was introduced to describe the wide spread, extensive manifestations of the disease in the jaws.¹ FCOD is a well recognized lesion predominantly affecting middle-to-old aged black women.² Once diagnosed, treatment is not generally necessary.² The cause of FCOD is still unknown, and there is no satisfactory explanation for the

reported gender and racial predilection. FCOD is defined as "Lobulated masses of dense, highly mineralised, almost acellular cemento-osseous tissue typically occurring in several parts of the jaw".² Although the 2nd edition essentially upheld the 1st edition's definition of FCOD, it modified the 1st edition's definition of periapical cemental dysplasia (PCD), another cemento-osseous dysplasia (COD), which most affected mandibular incisors to which may be adjacent to one another or in different parts of the jaws.² The problem with this is that it confuses the boundary between FCOD and PCD, if they are indeed two distinct lesions. The WHO definition only refers to the end-stage for FCOD, but

considers the natural history of PCD including initial radiolucent and final dense mineralized mass stages.² Furthermore, PCD need not be multiple, as the definition clearly recognizes that PCD can affect single teeth. The sole point that appears to separate FCOD and PCD is that for PCD, each periapical lesion is self-limiting, rarely exceeds 1 cm in diameter.² By creating a separate category of other cemento-osseous dysplasias for those lesions which share some of the features of periapical and/or florid cemento-osseous dysplasia, but do not have their characteristic clinicopathological patterns of presentation.² FCOD is a benign fibro-osseous lesion, typically involving multiple sites and frequently symmetric in distribution.¹ It is not unusual to find extensive lesions in all 4 posterior segments of the jaws and is a more extensive manifestation of the same lesion present in periapical and focal cemento osseous dysplasia.³ When the lesions are large, jaw expansion and facial deformity may be apparent. Radiographically, the lesions appear as multiple sclerotic masses, located in two or more quadrant, usually in the tooth-bearing regions.⁴ They are often confined within the alveolar bone.⁴ It is common to note these lesions in all four quadrants of the jaws. Involved areas of the jaw may undergo expansion. Histologically, the radiopaque lesions that are present appear to consist of an osteoid and cementum-like material, with fibroblasts being associated with the trabeculae of the calcified material.⁵ Simple bone cysts and inflammatory cells may also be associated with the lesions. Unless symptoms are noted, no treatment is normally rendered. Once infected the patient become symptomatic, and treatment of the secondary infection is very difficult, and antibiotics are often not effective.

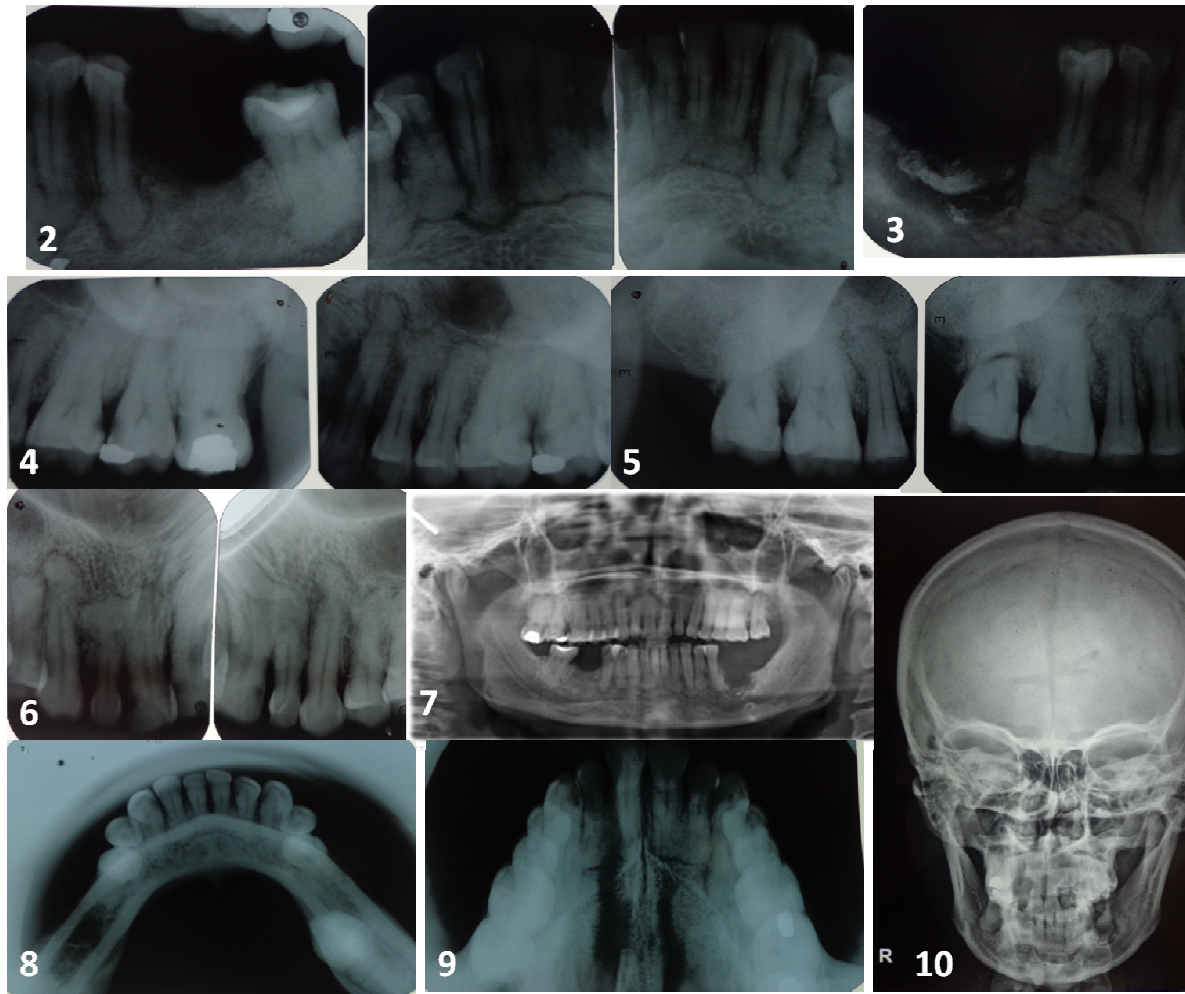
Case Report

A 45 years old female reported to the Department of Oral Medicine and Radiology with a complaint of pain in the left lower back tooth region of jaw since 1-2 months. History highlight that the patient felt pain when she got her tooth extracted in the same region 2 months back. Pain was severe and continuous and the extraction was traumatic as told by the patient. Extraoral examination highlight no significant changes. Intraoral examination revealed an unhealed socket with respect to 36, 37 region and patient was provisionally diagnosed as a case of Chronic Osteomyelitis with respect to 36, 37. (Figure 1)



Figure 1: Intra-oral Photograph showing unhealed socket w.r.t. 36, 37 region.

Intraoral periapical radiograph revealed loss of alveolar bone w.r.t. 36, 37 region and radioopacity w.r.t. periapical region of 35. Full mouth intraoral periapical radiographs were taken which revealed well circumscribed radiopacities surrounded with poorly defined radiolucency in both anterior and posterior region in both the jaws. (Figure 2-6) Panoramic radiograph revealed well circumscribed radiopacities surrounded with the poorly defined radiolucency in anterior and posterior region of both the jaws. Altered trabecular pattern was seen in these regions. Most of involved teeth showed hypercementosis. Masses appeared to be separated from root apices. (Figure 7)



Figures: 2-6: Intraoral periapical radiographs were taken which revealed well circumscribed radiopacities surrounded with poorly defined radiolucency in both anterior and posterior region in both the jaws; **7:** Panoramic radiograph revealed well circumscribed radiopacities surrounded with the poorly defined radiolucency in anterior and posterior region of both the jaws with altered trabeculae bone pattern; **8 and 9:** Maxillary and Mandibular occlusal view revealed well circumscribed radiopacities surrounded with the poorly defined radiolucency in anterior and posterior region of both the jaws; **10:** PA skull also revealed well circumscribed radiopacities surrounded with the poorly defined radiolucency in anterior and posterior region of both the jaws.

Maxillary and Mandibular occlusal view revealed well circumscribed radiopacities surrounded with the poorly defined radiolucency in anterior and posterior region of both the jaws. (Figure 8, 9) The PA skull also revealed well circumscribed radiopacities surrounded with the poorly defined radiolucency in anterior and posterior region of both the jaws. (Figure 10)

The blood chemistry showed that levels of serum alkaline phosphatase, calcium, Phosphorus, T3, T4, TSH, Parathormone all were within normal limits. Therefore, on the basis of Clinical, radiological and blood chemistry analysis, a diagnosis of Fcod with Chronic Osteomyelitis was given. A surgical intervention was sought; the infected area was opened, necrotic bone was removed and area was debrided. Necrotic bone was sent for histopathology

which further confirmed the diagnosis of Chronic osteomyelitis. (Figure 11)

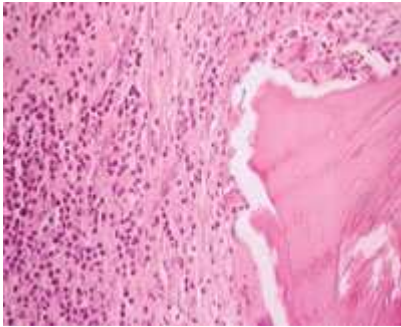


Figure 11: Histopathological photograph showing necrotic bone and inflammation.

Discussion:

Florid cemento-osseous dysplasia is a rare benign lesion arising from elements of the periodontal ligament and is strictly localized to the tooth bearing areas.⁶ Many lesions have to be differentiated from FCOD, and dental imaging can be used to distinguish between FCOD and other lesions that may exhibit a similar sclerotic appearance on conventional radiographs. Odontogenic tumors, especially cementoossifying fibroma, generally exhibit more buccolingual expansion than does FCOD.⁷ Dental imaging may be helpful in differentiating fibro-osseous lesions from odontoma, in which the CT number for enamel is higher than that for cementum. A screening dental panoramic radiograph is usually adequate for initial diagnosis. Radiographically the lesion varies depending on the stage. FCOD opacifies progressively as it becomes more mature. The classic appearance includes diffuse, lobular, irregular-shaped radioopacities throughout the alveolar process of the maxilla and mandible which was seen in our case.

Cementoblastoma characteristically is fused to the tooth apices. In hypercementosis, the cemental substance lies in continuity with the dental root, whereas in florid cemento-osseous dysplasia or gigantiform cementoma, it is separated from the periodontal space. This

latter feature is clearly evident in the radiograph of this patient.

Florid cemento-osseous dysplasia may have similarities with jaw bone changes in familial adenomatous polyposis (Gardner's syndrome)⁶, but florid cemento-osseous dysplasia has no other skeletal changes or skin tumours or even the dental anomalies that are seen in this syndrome as seen in our case. Another radiographic differential diagnosis is Paget's disease. The dysplastic lesions in Paget's are polystotic and the disease also shows biochemical serum changes. To rule out this dysplastic lesion serum alkaline phosphatase level were evaluated which were in normal range. Differential diagnosis of FCOD must also include sclerosing osteomyelitis, which can be a complication of the disease as seen in our case. However, etiopathogenesis is not clear.

The key points for this disease diagnosis, according to Brannon & Fowler⁸ are:

- Predilection for mid-age Black women;
- One or more circumscribed lesions (0.5 cm or shorter) at the periapical area of vital teeth;
- Painless non-expansive lesion located usually at mandible's anterior area;
- Radiographic characteristics can be radiolucency of mixed density (radiolucent with opacities), or opaque with a narrow radiolucent margin;
- Cellular fibrous stroma with lamellar osseous tissue and/or oval calcifications.

The case here described fulfills almost all the characteristics of the diagnosis suggested by Brannon & Fowler. This case was unusual as there was presentation of concomitant osteomyelitis and FCOD. The management of FCOD is twofold. In asymptomatic patients it is probably wise to keep the patient under observation without surgical intervention. Because the onset of symptoms is usually associated with exposure of the sclerotic masses to the oral cavity, biopsy or

elective extraction of teeth in the involved area should be avoided. This can lead to bone necrosis, which may be treated difficulty conservatively with antibiotic therapy.⁵ At this stage, there is an inflammatory component to the disease and the process is basically a chronic osteomyelitis involving dysplastic bone and cementum as seen in our case.

Saucerisation of dead bone and cementum is the treatment recommendation in the literature as suggested to our patient. It would be impractical to completely resect the lesion because it usually occupies most of the mandible and maxilla. When surgical intervention is indicated, a remodeling resection is recommended for esthetic reasons. When the patient is symptomatic secondary to a tooth pain, the tooth may be managed endodontically by avoiding extractions. Cases with secondary predisposed factor of infection are difficult and complicated to manage.

Conclusion

This report clearly highlights a rare case of FCOD. Most cases of florid cemento osseous dysplasias are asymptomatic and are found during routine radiographic examination. No surgical interventions are indicated unless it is symptomatic because it can lead to secondary infections which are difficult to treat.

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Source of support: Nil

Conflict of interest: None declared