Florid Cemento-Osseous Dysplasia at the Site of Previous Teeth Extraction: Report of a Case

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Abstract

Objective: Florid cemento-osseous dysplasia (FCOD) is a rare bone lesion that predominantly involves the women’s jaws in middle age. This condition is usually asymptomatic and has a benign course.

Case: This paper presents a rare case of FCOD in a white middle aged woman, which had affected mandible bilaterally and was diagnosed after tooth extraction and treated conservatively. We believed tooth extraction was a contributing factor for outbreak of such a lesion in this susceptible patient.

Conclusion: For the asymptomatic patients, the best management consists of regular recall examinations with prophylaxis and reinforcement of oral hygiene to prevent periodontal diseases and tooth loss, but with accession of clinical signs and symptoms, surgical intervention is inevitable.

Key words: Bone diseases, Florid cemento-osseous dysplasia, Fibrous dysplasia of bone, Osteomyelitis, Tooth extraction, Mandible.

Please cite this article as: Akhlaghi F, Bemanali M, Dehghani N. Florid cemento-osseous dysplasia at the site of previous teeth extraction. J Dent Sch 2015; 33(2): 175-181.

Received: 02.09.2014          Final Revision: 26.04.2015          Accepted: 04.05.2015

Introduction:

The fibro-osseous lesions (FOL) are kind of bone pathologic conditions in which fibrous connective tissue replace normal bone (1). Cemento-osseous dysplasia, which is the most common type of fibro-osseous lesions, commonly arises from periodontal ligament fibroblasts and like other fibro-osseous lesions, normal bone gets replaced with fibrous connective tissue containing abnormal bone or cementum-like tissues. Florid cemento-osseous dysplasia is a dysplastic or developmental bone lesion of the jaws. It tends to affect middle aged black women, although it has been also seen in other races and males. In some cases, a familial tendency has been reported. The symptoms of the lesion vary extensively from asymptomatic lesions which are only detectable in casual radiographs, to symptoms such as dull pain, drainage, and infection signs which are almost always associated with exposure of sclerotic calcified masses in the oral cavity. This exposure of pathologic mass can take place iatrogenically after tooth extraction in the involved jaw’s area or may be a consequence of progressive alveolar ridge atrophy as the senile process or prolonged denture rehabilitation (2-5).

The radiographic appearance of the lesion is well-defined as mixed radiolucent-radiopaque with radiolucent periphery and surrounding sclerosing border. These multiple sclerotic lesions involve multi quadrants in jaws, particularly in posterior mandible and above the mandibular canal (the tooth-bearing areas) (6). Histologically, these lesions are composed of
compact, a cellular cemetum or osseous-like tissue embedded and surrounded by peripheral rims of fibrous connective tissue (7). This paper reports a case of FCOD in a patient who was presented after tooth extraction and describes clinical and radiographic features of this lesion and discusses its differential diagnosis and etiologic factors.

**Case Report:**

In April 2003, a 38-year-old white female presented for dental care. Her past medical history was clear. On physical examination, she had no extra-oral pathologic signs and symptoms (Figure 1).

Intra-oral examination indicated a partially edentulous area in right posterior mandible and the gingival and buccal mucosa of that region were normal without any clinical signs of inflammation (Figure 2A).

Radiographic examination showed multiple sclerotic masses with radiolucent borders in mandible (Figure 3).

**Figure 1- Extra-oral view of the patient**

**Figure 2- Intra-oral view of the patient. A, Right mandibular quadrant. B, Left mandibular quadrant.**

**Figure 3- Panoramic radiograph showing multiple well-defined sclerotic masses with radiolucent border in right molar region of the mandible**
Two years before that in 2001, she had had her right mandibular first molar extracted due to toothache. In 2005 follow up visit, she had no dental problem and radiographic examination was showed the same intra-bony masses with more sclerotic changes (Figure 4).

After 4 years in 2009, she presented for periodontal disease at the left mandibular second molar which had to be extracted. We observed again a multiple sclerotic mass in the left extraction site in the panoramic radiograph and in the next years’ follow up visits which her radiographs showed the same process (Figure 5). During 10 years follow-up the patient had been totally asymptomatic and the lesion had been detected when radiographs were taken for some other purposes, so it was not necessitated to surgical intervention (Figure 2B, 6).

Considering that FCOD is usually diagnosed clinically and radiographically, the biopsy of the lesion is not recommended. Biopsy can be accompanied by the risk of jaws fracture or infection. In this case, we decided not to surgically excise the lesion because of its asymptomatic feature and considering that surgical intervention for such a lesion requires an aggressive surgery and may leave extensive defects in jaws.
Discussion:

Classically, cemento-osseous dysplasia is categorized into three subgroups according to World Health Organization (WHO) calcification:

1. Periapical cemento-osseous dysplasia: lesions in the anterior mandible and only a few adjacent teeth.
2. Focal cemento-osseous dysplasia: limited number of lesions in mandibular posterior teeth-bearing or edentulous areas.
3. Florid cemento-osseous dysplasia: more extensive forms, occurring bilaterally in the mandible or all jaw quadrants (8).

Radiographic feature of cemento-osseous dysplasia has a wide spectrum in appearance from radiolucent or mixed radiolucent/radiopaque lesions in early stages to radiopaque lesions in late stages; depending on its maturation from fibrous tissue to calcified cementum-like tissue (9).

Histopathologically, this lesion is a vascular, fibrous connective tissue, which is a benign fibro-osseous lesion that must be differentiated from other benign fibro-osseous lesions such as fibrous dysplasia, ossifying fibroma, Paget’s disease, gigantiform cementoma, enostosis or...
exostosis and sclerotic osteomyelitis on the basis of combined clinical, radiographic and histological features (10, 11).

Fibrous dysplasia can occur in any part of the skeleton and affect one or several bones. In skull, maxilla is the dominant site. Clinical manifestation of this disorder is usually evidenced in childhood and it has no gender or race predilection (12). Ossifying fibroma is also prevalent before adolescence. It affects often the maxilla, particularly premaxilla regions around cuspid teeth (13).

Paget’s disease is a chronic progressive disorder of the bone that can result in abnormal bone architecture. Often this condition is localized to only a few bones in the skeleton and spine, femur, skull, pelvic and sternum are the most commonly affected bones. In skull region, mandible is commonly affected and the entire jaw is involved with loss of lamina dura, whereas florid cementos-osseous dysplasia is centered above the inferior alveolar canal. Paget’s disease is caused by the excessive bone breakdown and formation, followed by disorganized bone remodeling which often results in biochemical serum changes, especially elevated alkaline phosphate levels; in contrast to cemento-osseous lesions that are not accompanied with biochemical serum changes (14-16).

Florid cemento-osseous dysplasia is sometimes misdiagnosed as chronic sclerosing osteomyelitis, particularly when it is symptomatic such as lesions accompany pain and discharge. However, sclerosing osteomyelitis usually affects the mandible and manifests clinical signs of infection like pain, swelling, fistula development and pus formation and shows radiographic signs of diffuse sclerosis and ill-defined osteolytic areas; whereas florid cemento-osseous dysplasia indicates mixed radiolucent/ radiopaque lesions with multiple lobulated opaque masses. Sclerosing osteomyelitis involves the mandible generally, which can spread from the alveolus crest to the inferior border of body and in severe cases, its extension to the ramus is conceivable. Also, this condition occurs predominantly in adult men. In contrast, FCOD often involves tooth-bearing areas and has a tendency to involve black women (2, 7, 17-19).

FCOD may be similar to changes of jaw related to familial adenomatous polyposis (Gardner’s syndrome), although it does not have other presentations of Gardner’s syndrome such as polyps gastrointestinal tract, skull ostemas, skin tumors or dental anomalies. Although some authors have reported cases of FCOD with familial inheritance pattern of autosomal dominant or in some other cases with unknown pattern of genetic transmission, but this is not a common feature for this lesion (20).

Another differential diagnosis for FCOD is familial gigantiform cementoma (FGC), which belongs to category of the fibro-osseous lesions and thus creating great confusion in the differential diagnosis. FGC is rare benign tumor of the jaws with autosomal dominant genetic pattern which occurs mostly in children without any gender predilection. The lesion typically tends to affect multiple quadrants in both jaws with considerable expansion and potential for producing asymmetry and facial disfigurement. Whereas, the FGC has a neoplastic behavior, and surgical intervention for treatment is inevitable (21).

Enostosis and exostosis sometimes have similar radiologic features with FCOD and place as differential diagnoses for this lesion. Exostosis develops on the surface of the jaws as the result of new bone formation with normal bone pattern. Its common location is on buccal surface of posterior maxillary teeth which is palpable in touching the mucosal surface. Radiographically, it shows highly dense radiopacities in tooth bearing areas of the jaws without any radiolucent borders or compartments. On the other hand, enostosis is a distinct island of
Fibrous dysplasia

cortical bone within the cancellous bone without any clinical signs or symptoms and may be only identified incidentally on radiographs. Like exostosis, they have pure radiopaque features in radiographs without a radiolucent rim surrounding the radiodense area or mixed radiopaque/radiolucent view, unlike FCODs (22).
The etiology of the cemento-osseous dysplasia is not well-established; however periodontal ligament origin is the most common accepted hypothesis. It is believed that fibroblastic mesenchymal stem cells, which are in the periodontal membrane, proliferate and develop such lesions. These stem cells are cementoblastic precursor cells (2). Some authors suggest trauma, dental caries, periodontal disease, or infection as stimulating factors for developing such a lesion (23, 24). In this presented case, tooth extraction can be considered as a contributing factor for initiating of this pathologic process. We can conclude that trauma in any form, either evident like this case which was the result of tooth extraction, or unremarkable trauma can be triggering factors for outbreak of these lesions in susceptible patients. In addition, the periodontal ligament origin of these lesions can be clarified in such a case and strengthen this hypothetic origin role of periodontal ligament cells.
Normally, a diagnosis of florid cemento-osseous dysplasia in the jaws is made by clinical findings, radiographic features, considering the site of lesion development, age, gender and ethnicity of the patient. Asymptomatic cases need no treatment because the lesion is self-limiting. Management of these conditions includes clinical and radiological follow up. These follow-up examinations are recommended for a minimum of two years. Once a patient becomes symptomatic, surgical intervention and antibiotic therapy are inevitable. Management of secondary infection may be complicated because the involved bone cannot diffuse the antibiotics properly, so surgical debridement of infected cemento-osseous-like structures is needed (11, 20, 23).

Conclusion:

FCOD is a rare condition of the jaws bone, commonly seen incidentally in routine radiographic examinations and confirms diagnosis by its characteristic clinical and radiologic features. The etiology of FCOD is not well recognized and some contributing factors for outbreak of this lesion are mentioned such as trauma, caries, infection and periodontal disease. In this presented case we shed light on the effect of trauma in the form of tooth extraction as the initiating factor for this lesion.

Conflict of Interest: “None Declared”

References: