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Florid Cemento-Osseous Dysplasia: Report of Three Symptomatic Cases

Florid cemento-osseous dysplasia (FLCOD) is a fibroosseous lesion that affects the jaw of middle-aged women. This lesion occurs more frequently in Black and Asians. A familial trend has been reported for some cases. This lesion may be completely asymptomatic and in such cases the lesion is detected in the radiographs incidentally. Radiographic features include multiple radiopaque masses, usually located in three or four quadrants or can be extensive throughout one jaw. However, when they are present in only one jaw, the mandible is the more common location. In this paper we report three cases of FLCOD that had evidences of infection as the first presentation.

Keywords: Florid Cemento-Osseous Dysplasia, Florid Osseous Dysplasia, Radiography, Panoramic

Introduction

Florid cemento-osseous dysplasia, previously called gigantiform cementoma, multiple cemento-ossifying fibroma, sclerosing osteitis, multiple enostosis, and sclerotic cemental masses of the jaws, was first described comprehensively by Melrose et al. The etiology of this disorder is unknown. Histopathology shows it may be the result of reactive or dysplastic changes of the periodontal ligament.[1]

Florid cemento-osseous dysplasia is more commonly seen in middle-aged black women, although it may also occur in Caucasians and Asians. In some cases, it may be familial. The lesion may be completely asymptomatic and occasionally it is detected incidentally when radiographs are taken for some other purposes. Symptoms such as dull pain or drainage are almost always associated with the exposure of sclerotic calcified masses in the oral cavity which may occur secondary to progressive alveolar atrophy under a denture or after extraction of teeth in that region.[2]

Radiographically, the lesions appear as multiple sclerotic masses located in two or more quadrants usually in the tooth-bearing regions often confined within the alveolar bone. The lesions typically demonstrate two identical patterns of maturation; initially, the lesions are predominantly radiolucent but become mixed gradually, then dominantly radiopaque with only a thin peripheral radiolucent rim. Occasionally, a lesion may become almost totally radiopaque and blend with the adjacent normal appearing bone.[3]

Large FLCOD lesions can displace the inferior alveolar nerve canal downward. FLCOD may also displace the floor of the antrum upward and may cause enlargement of the alveolar bone by displacement of the buccal and lingual cortical plates. Extraction of the associated teeth may be difficult because of the remarkable hypercementosis of them that fuses with the abnormal cemental tissue of the lesion.[4]

Histologically, these lesions have a fibroblastic background, which consists of anastomosing bone trabeculae and multiple layers of calcifications.[2]

In most instances of FLCOD, the distinctive clinical and radiographic patterns (i.e.,

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a black female with multiquadrant involvement) allow a strong presumptive diagnosis without the necessity of biopsy.[3]

Radiographic evaluations should be used for follow-up.[2] This paper describes three white women who presented with infection and were diagnosed with florid cemento-osseous dysplasia on the basis of clinical, radiographic, and histological findings.

FLCOD diagnosis is valuable because the bone density is increased and bone vascularity is decreased which predispose osteomyelitis of the bone. Hypercementosis is another risk factor for traumatic tooth extraction. The above mentioned factors promote to increase the the risk of osteomyelitis in these patients.

We report a very rare case of CCF who presented with unilateral blindness.

Case Presentation

Case 1

A 45-year-old woman suffered from pain in the left side of the mandible since three months ago. She had pus drainage from the left posterior segment of the mandible and had not been able to use her denture.

In the panoramic view, a well-defined radiopaque lesion without a radiolucent rim in the right posterior region of the mandible and another well-defined radiopaque lesion were seen in the left side of the mandible, which had a radiolucent capsule probably due to infection. Finally, the diagnosis of FLCOD was established (Fig. 1).

Case 2

A 52-year-old woman reported a swelling in the right posterior maxilla and posterior mandible since many years ago. She also had halitosis and pus drainage from the swollen regions. She reported occasional increase and decrease in the size of the lesions. Clinical examination revealed a bony hard expansion in the anterior vestibule of the mandible and the posterior segment of the right maxilla.

In the panoramic view, multiple well-defined opaque lesions with a radiolucent capsule were seen extending from the anterior segment of the mandible to the third right mandibular molar. These lesions were located in the apical region of those teeth and above the inferior alveolar canal. Several hypercementoses were also seen in some of them. The same lesions were evident in the anterior and left posterior segment of the maxilla. Eventually, the radiographic findings confirmed the diagnosis of FLCOD (Fig. 2).

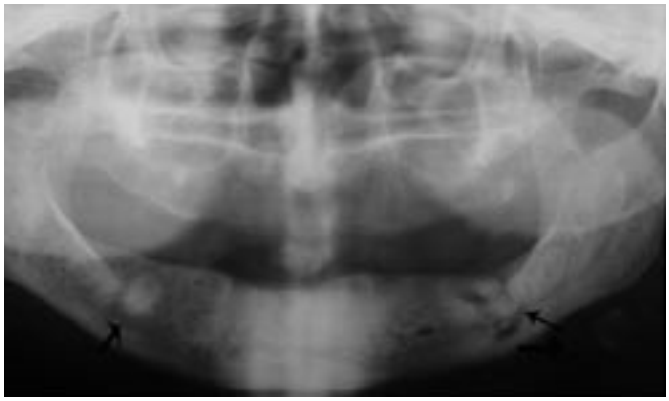

Fig. 1. A 45-year-old woman with mandibular pain and pus drainage. The panoramic view shows two well-defined radiopaque lesions in both quadrants of the mandible.

Case 3

A 57-year-old woman reported a swelling in the region of the left maxillary incisors from three months ago. She had not been able to use her denture since

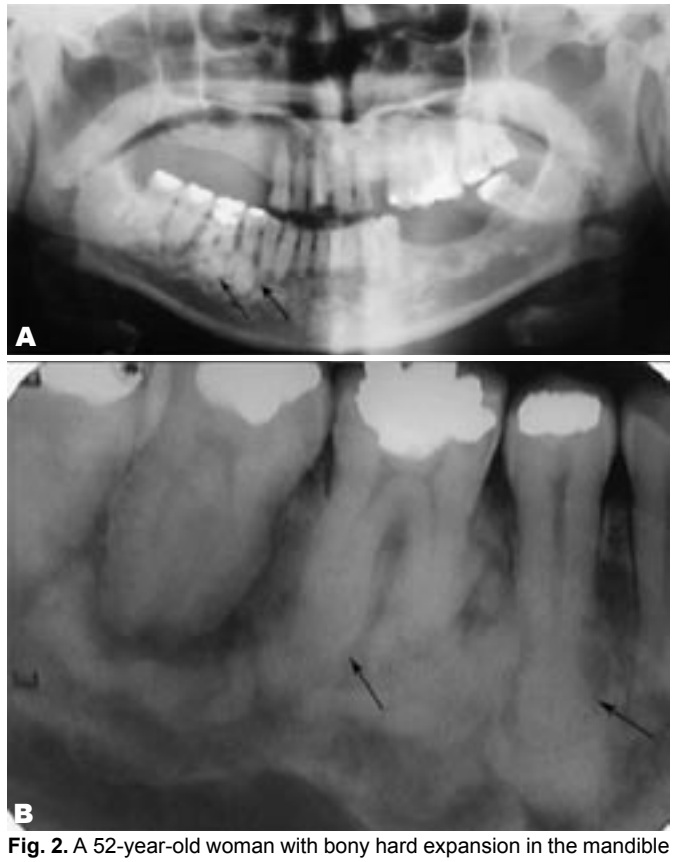

Fig. 2. A 52-year-old woman with bony hard expansion in the mandible and maxilla.
A. Panoramic view indicating multiple radiopaque lesions.
B. Periapical view demonstrating several hypercementoses in the roots of the teeth.

1 month ago. She had mild pain together with pus drainage in the morning, We found an expansion in the region of mandibular incisors and the right maxillary canine.

In the panoramic view, multiple opaque lesions with a radiolucent rim in the anterior mandibular region, anterior segment of the maxilla and in the first left maxillary molar were seen. Finally, the diagnosis of FLCOD was established (Fig. 3).

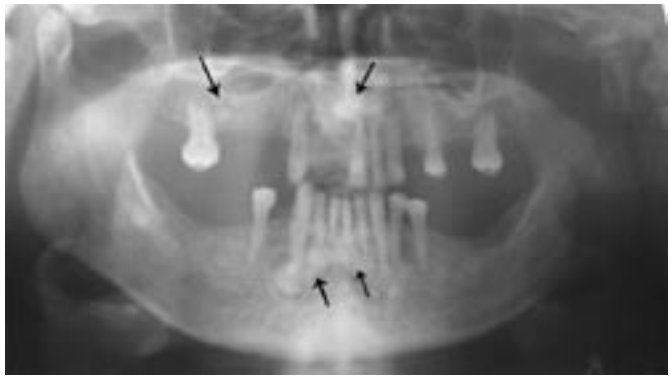

Fig. 3. CT angiography with 3D VRT images revealing giant aneurysm in the cavernous portion of the left internal carotid artery.

Discussion

FLCOD has been described as a dysplastic lesion or developmental anomaly arising in tooth-bearing areas. FLCOD typically occurs in middle-aged black Asian women. Melrose et al.[5] reported a study of 34 cases of such lesions, of which 32 were black women (in a predominantly Caucasian population) with a mean age of 42 and Mac-Donald[6] also reported sixteen cases of FLCOD (mean age, 52.1 years) of which six were West Indian women and ten were Chinese.

In this study, all three cases were middle-aged Caucasian women. Although these lesions are often asymptomatic and may present as incidental radiological findings, in our study, there was mild pain and pus drainage in all the cases. Symptoms may be due to secondary infection of exposed masses.

A rare type of FLCOD is aggressive and forceful surgical interventions may be required. None of our cases had the aggressive type of FLCOD.[1]

Radiologically, FLCOD consists of masses with different degrees of opacity with or without a radiolucent margin. The diagnostic criteria include diffuse alveolar involvement not limited to the apices of the teeth and affecting more than one quadrant.[4,5] Radiological findings of the present case concur with these criteria, and in one of our cases only the mandible was involved, but in the other cases both jaws were involved. The roots of the associated teeth may have a considerable amount of hypercementosis[4] similar to one of our cases that had a large amount of hypercementosis in her teeth.

Radiologically, it must be differentiated from other lesions such as Paget's disease, sclerosing osteomyelitis, periapical cemento osseous dysplasia (PCOD), focal cemento-osseous dysplasia (FCOD) and multiple idiopathic osteosclerosis. PCOD and FCOD have several similarities with FLCOD including age, sex and racial profiles of the patients and comparable radiographic and histologic appearances. However, FCOD is a single lesion compared with FLCOD that consists of multiple lesions. PCOD usually involves the anterior segment of the mandible, but FLCOD usually involves three or four quadrants.[4]

Idiopathic osteosclerosis is a well-defined lesion without a radiolucent rim that has a bony internal structure, whereas FLCOD has a cemental internal pattern. Idiopathic osteosclerosis also occurs as multiple lesions in patients with colorectal carcinoma or adenoma and Gardner syndrome.[7]

Paget's disease of the bone may have a cotton-wool appearance. However, this condition affects the bone of the entire mandible, while florid cemento-osseous dysplasia is placed above the inferior alveolar canal. Paget's disease shows loss of lamina dura, but lamina dura is not affected by FLCOD.[8] Paget's disease is often polyostotic, involving other bones such as the spine, femur, skull, pelvis and sternum[4] and leads to biochemical serum changes, such as elevated alkaline phosphatase levels.[9] No involvements of other bones were found in these reported cases.

Chronic diffuse sclerosing osteomyelitis is another disease that should be differentiated from FLCOD. It appears as a single, poorly delineated opaque segment of the mandible, whereas florid cemento-osseous dysplasia is seen as multiple round or lobulated opaque masses. Chronic diffuse sclerosing osteomyelitis involves the body of the mandible.[2,10,11] In addition, FLCOD is frequently associated to black women, while chronic diffuse sclerosing osteomyelitis does not generally occur in women.[10,11]

Florid cemento-osseous dysplasia may be familial; however, no familial aspects of the disease could be

established in our cases.

In conclusion, generally when a lesion is found in the jaws, the diagnosis is suggested by clinical and radiographic findings and is confirmed by histopathology. However, FLCOD is a condition in which the diagnosis relies on radiology and clinical findings alone, and biopsy is not mandatory due to the increased risk of infection.

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