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Case Report

Co-Occurrence of Florid Cemento-Osseous Dysplasia and Chronic Diffuse Osteomyelitis

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ABSTRACT

Florid Cemento-Osseous Dysplasias are benign, lesions. However, exposure of the sclerotic masses to the oral cavity may lead to secondary infection, which may be difficult and complicated to manage. Since the radiographic and histologic spectrum is similar to other fibro-osseous lesions with a different treatment plan, an accurate diagnosis is necessary. Surgical intervention is not the optimal treatment, as it may result in secondary complications; in fact, patient is kept under frequent follow-ups. The present case describes florid cemento-osseous dysplasia revealed by chronic diffuse osteomyelitis.

Keywords: Florid cemento-osseous dysplasia, osteomyelitis, diagnosis, treatment.

INTRODUCTION

Florid Cemento-Osseous Dysplasia (FCOD) is one of the subgroups of cemento-osseous dysplasia (COD) which was first described by Melrose, Abrams and Mills in 1976. [1,2,3,4,5,6]

It is more commonly seen in middle aged black women although it may also occur in Caucasians and Asian. The etiology of FCOD is unknown, although, some authors have proposed that a reactive or dysplastic process of the periodontal ligament may be involved.

The processes may be totally asymptomatic and may be detected incidentally when radiographs are taken for some other purposes. However, other symptomatic cases are almost associated with dull pain or drainage and exposure of sclerotic calcified masses in the oral cavity. For these symptomatic cases, FCOD is often misinterpreted as chronic sclerosing osteomyelitis, which can be a complication associated with this entity. [1,3,5,6,8,9]

CASE REPORT

The aim of this article is to report clinical case of FCOD which be treated at the department of Medicine and Oral Surgery of the Dentistry Clinic of Monastir, Tunisia.

A 79 year-old woman with no serious systemic disease was referred by her general dental practitioner to our department. Extra oral examination revealed diffuse swelling of the mandible sensitive on palpation (figure 1).

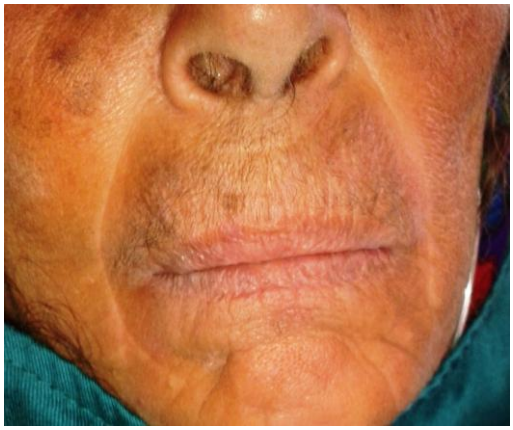

Figure 1: Bilateral swelling on the jaw.

There was no mandibular nerve hypoesthesia. An intraoral examination showed poor hygiene, plaque and calculus accumulation and gingival inflammation. The examination of posterior mandibular area show an erythematous oral mucosa and multiple productive fistulas of purulent exudate upon palpation at the bilateral posterior mandibular edentulous ridges (figure 2).

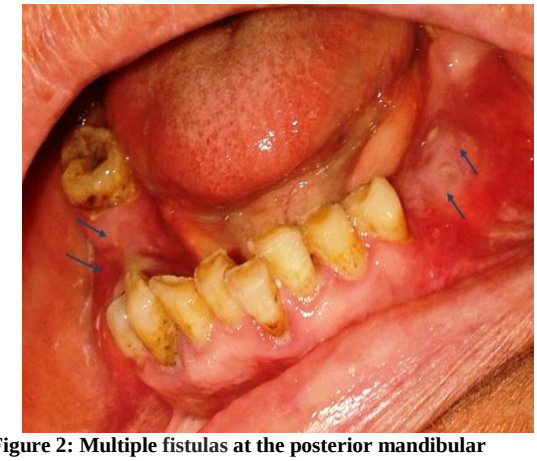

Figure 2: Multiple fistulas at the posterior mandibular edentulous ridges.

Panoramic radiograph showed ovoid radiopaque masses surrounding roots of molars close of wide radiolucent spaces in both quadrants of the mandible. Lesions are unconnected with the mandibular canal (figure 3).

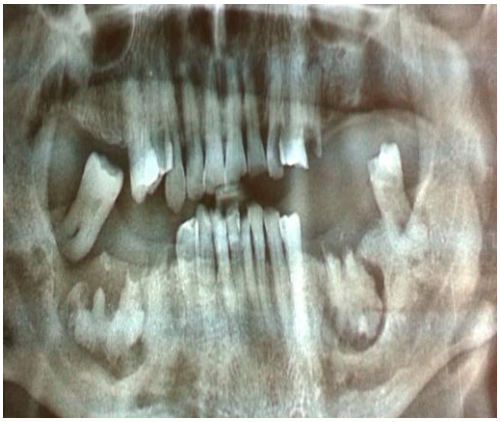

Figure 3: Panoramic radiograph showing radiodense/ Radiolucent lesions closely associated with the roots of molars mandibular.

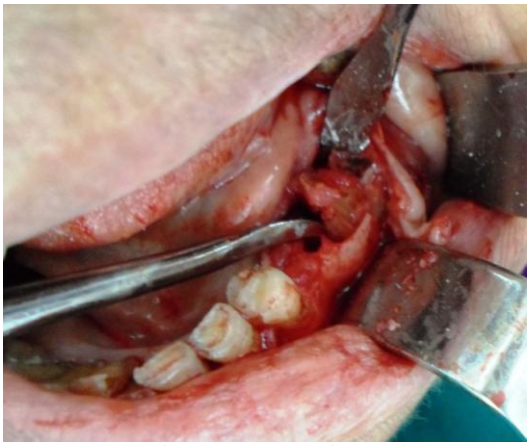

Figure 4: Excision of hard masses in left side of mandible.

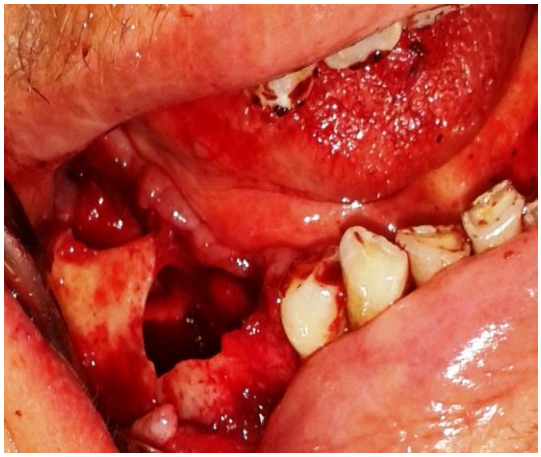

Figure 5: Operative cavity following to the lesion removal in right side of mandible.

Clinical and radiographic signs may evoke infected florid cemento-osseous dysplasia; osteitis or cementomas.

The patient was then treated with antibiotics to eradicate the infection, after, two surgeries were performed to excise hard mass and to curettage of granulation tissue under local anesthesia (figures 4, 5, 6, 7).

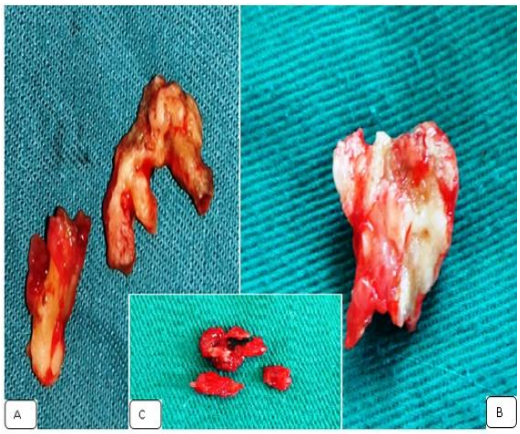

Figure 6: specimens. **A: right side; B: left side; C: tissue of granulation.**

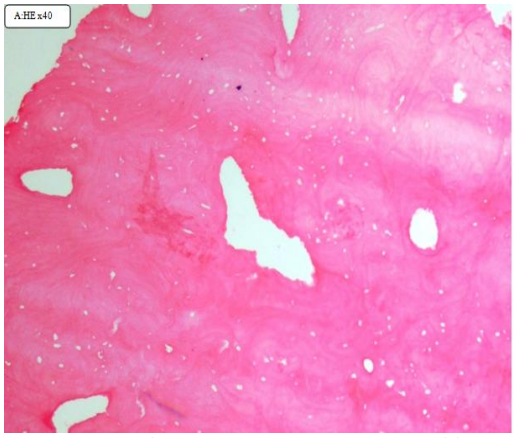

Figure 7: Control after a week and disappearance of fistulas.

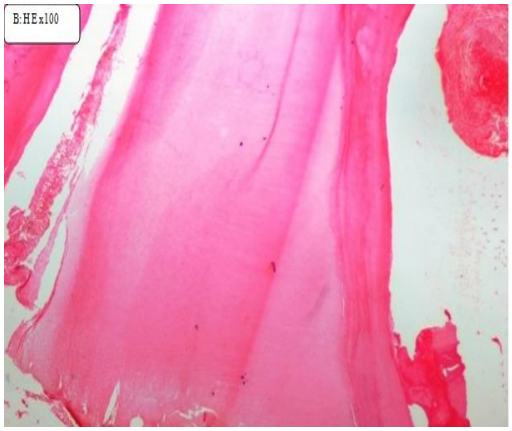

Figure 8: Anatomopathological examination of the lesion showing an irregular mass of cemento-osseous tissue within a fibrous stroma.A: H.E ×40, B: H.E ×100.

The specimens were subjected to an anatomopathological examination. The results revealed a mass of cemento-osseous tissue within a fibrous stroma (figure 8).

The diagnosis of florid cement-osseous dysplasia complicated by chronic suppurative osteomyelitis was thus made.

DISCUSSION

COD lesions are classified by World Health Organization in 2005 depending on their topography and radiographic appearance, into three main groups: periapical, focal and florid cemental. They are considered as bone related non neoplastic fibro-osseous lesion in which the normal cancellous bone is replaced by dense, acellular cemento-osseous tissue in a background of fibrous connective tissue. [10]

FCOD is the most rare variant of the COD lesions. The term "florid" refers to its excessive and widespread location occurring bilaterally in the mandible or in all jaw quadrants. [1]

Radiographically, FCOD appears as dense, lobulated masses and often symmetrically located in various regions of the jaws. Theses lesions have three developmental stages which have different radiographic images. First, osteolytic stage is seen as well defined radiolucent area with loss of lamina dura and periodontal ligament when lesions are associated to teeth. The second cement blastic stage, small radio opacities appears in radiolucent area, because of deposition of cementum like droplets in fibrous tissue. Last stage is described as definite radio opacity present in majority of the lesion. [2]

The diagnosis is made by clinical and radiographic examination. If they are enough to diagnose, biopsy should be avoided. Otherwise, an antibioprophylaxis is indispensable, previous to biopsy because the risk of infection, sequestrum formation and osteomyelitis risks, due to the poor

socket healing from the impaired blood circulation in the affected area of the bone. [2]

Differential diagnosis of FCOD must be made with Paget's disease of bone, Gardner's syndrome and chronic diffuse osteomyelitis. All these pathologies have in common lesions of the jaws. Thus Gardner's syndrome can be differentiated from FCOD by skeletal change, skin tumors, and dental anomalies. For Paget's disease is polyostotic and shows the raised alkaline phosphatase level which is not a consistent feature of FCOD. Concerning chronic diffuse osteomyelitis, is an inflammatory condition of the mandible presenting with cyclic episodes of unilateral pain swelling and shows a single area of diffuse sclerosis containing small, ill defined osteolytic areas, associated with long standing infected tooth. Whereas FCOD is seen as multiple round or lobulated opaque masses but in rare case osteomyelitis can be a complication associated with FCOD disease. Thus, pain, drainage, exposure of the lesion in oral cavity, focal expansion and facial deformities are present. [1,5,11,12]

Treatment is not required for asymptomatic cases of FCOD. Indeed, the patient should be regularly follow-up to detect behavioral change of the lesions and panoramic radiographs in every 2 or 3 years is recommended with prophylaxis and reinforcement of hygiene care to control periodontal disease and prevent tooth loss. An antibioprophylaxis is recommended prior to endodontic treatment or tooth extraction in FCOD affected patients, to avoid the risk of infection.

Management of the symptomatic patient is more difficult because chronic inflammation and infection develop within densely mineralized tissue. Treatment options of these patients are antibiotic therapy and sequestrectomy. The treatment of a secondary infection of this lesion can be difficult and complicated. [4,5,13]

The progressive deposition of cementum, lesion's maturation in COD, increases the risk of secondary osteomyelitis. This deposition may occur because of poor vascularity of the altered dysplastic tissue when there is communication with the oral cavity. Kawai and colleagues reported that COD was accompanied by osteomyelitis in 14.8% of their 54 cases. Groot and colleagues reported that diffuse sclerosing osteomyelitis occurred in association with florid COD in 31.3% of their 16 cases. Noura and colleagues reported in their study that13 (11.0%) of 118 patients with COD presented with concomitant osteomyelitis.

Although they affect only a small number of patients, the potential for complications of this type to arise because of exposure through tooth extraction sites, bone loss from periodontal disease or implant placement should be recognized. [14]

To conclude, the diagnosis of FCOD, which can include several different but related entities, should consider tooth pulp vitality, the stage of lesion development and the possible co-existence of FCOD with other entities, like osteomyelitis. In fact osteomyelitis and FCOD are two independent disease processes that may have overlapping clinical and radiographic characteristics. Differential diagnosis can be crucial, as the course of each process and its clinical management varies. [5,14]

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