ABSTRACT: Florid osseous dysplasia (FOD) is an extensive form of osseous dysplasia where normal trabecular bone is substituted by fibrous connective tissue and amorphous mineralized tissue. Usually, the lesions are mainly asymptomatic and the patients should be followed with clinical and imaging examination, requiring no intervention. Nevertheless, due to the poor vascularization of the lesion and to local trauma, secondary infections and osteomyelitis may occur. Patients may present with pain, mucosal ulceration, lesion exposure in the oral cavity, fistula and swelling. In such cases, the correct diagnosis and management of the lesion is decisive to reestablish patient’s health and quality of life. The aim of this article is to present a case of complicating secondary chronic osteomyelitis treated successfully with conservative intervention. A 68-year-old black female patient reported a “swelling of the gums” that was present for eleven years, with episodes of periodic pain and swelling. On physical examination, a papule with suppuration in the alveolar mucosa in the right side of the mandible was observed. Panoramic radiography and CBCT showed a mixed lesion surrounded by sclerotic bone. The patient was diagnosed with chronic osteomyelitis in association with FOD and treated with antibiotic therapy together with surgical curettage. The incidence, etiophatology, differential diagnosis, treatment and prevention of secondary osteomyelitis associated with FOD are discussed in the light of literature. This information might assist the dentists while choosing the best treatment options for similar cases.

KEY WORDS: displasia cemento-ósea florida, osteomielitis.

INTRODUCTION

Benign Fibro-Osseous Lesions (BFOL) are a group of conditions with similar histological features where normal bone is replaced with a fibrous connective tissue with different degrees of mineralization. Each BFOL subtype presents unique demographic, clinical and radiologic characteristics (de Noronha Santos Netto et al., 2013). Osseous dysplasias (OD) are the most common BFOL of the jaws (de Noronha Santos Netto et al.; Abramovicz & Rice, 2016). The World Health Organization (WHO) refers to these conditions as "osseous dysplasia" and recognizes the term "cementum-osseous dysplasia" as a synonym (Barnes et al., 2005).

OD have been designated according to their features, as periapical, focal, florid or familial gigantiform cementoma (FGC). Florid osseous dysplasia (FOD) was first described by Merlose et al. (1976) as an exuberant form of OD characterized by the presence of multiple, diffuse, lobular or irregular-shaped radiopacities throughout the alveolar processes, but not restricted to the root apices. FOD has a remarkable tendency to bilateralism, usually, with symmetrical lesions and can involve all quadrants (Melrose et al.; Barnes et al.; Sadda & Phelan, 2014). Simple bone cists concomitant occurrence had also been reported. Cortical bone expansion may be present, however external visible facial asymmetry is usually not observed (Melrose et al.).

Characteristically, FOD affects middle-aged black females (Barnes et al.). In the majority of the cases, no symptoms are present and the lesions are identified when radiographs are taken for another
The purpose (Barnes *et al*.; Alsufyani & Lam, 2011). The diagnosis of FOD should be carried out with clinical and imaging evaluation, which provides essential information for the correct selection of treatment (MacDonald-Jankowski, 2004). In fact, biopsy should be avoided in asymptomatic lesions because of the risk of infection and fracture. The surgical removal of the lesions is also not justified since it would involve extensive intervention and FOD lesions are self-limiting (Dagistan *et al*., 2007; Alsufyani & Lam; Kutluay Köklü *et al*., 2013).

FOD management depends on the presence of symptoms and no treatment is required unless the lesion becomes secondary infected. When symptoms of infection are present, aggressive curettage should be done, removing the dead tissue. In Asymptomatic lesions clinical and radiographic follow-up should be carried (Sadda & Phelan). The aim of this article is to report a case of conservative treatment with antibiotics and curettage of a secondary chronic osteomyelitis in a patient with FOD.

CASE REPORT

A 68-year-old black female patient with a complaining of "swelling in the gums" was referred to the Department of Surgery, Pathology, Radiology and Stomatology at Bauru School of Dentistry. The lesion was already present 11 years ago, with episodes of periodic pain and swelling. The patient also reported having undergone three previous surgical interventions in the jaws. Her medical and family history were not remarkable. The intraoral examination revealed the presence of a 1 mm of size papule, bleeding when touch and showing suppuration in the alveolar mucosa in the right side of the mandible. The patient was toothless and had been using tissue-borne prosthesis (Fig. 1).

An antibiotic therapy was carried with clindamycin 300 mg, four times, daily. After seven days of antibiotic therapy the surgical intervention was performed. Under local anesthesia the necrotic bone was removed by surgical curettage. The oral surgical soft tissue was submitted to histopathologic examination. The examined section features correlated with fistula containing intense chronic inflammatory infiltrate. The antibiotic prescription was maintained for 5 days after surgery. The patient has been followed-up and no sign of recurrence have been observed during one year.

A panoramic radiography showed multiple irregular and diffuse radiodense masses in maxilla and mandible (Fig. 2). In the posterior region on the right side of the mandible, a radiopaque and radiolucent lesion was observed (arrow). The radiographic images correspond to the area of intraoral fistula.

Cone beam computed tomography revealed mixed hypodense/hyperdense lesion with discontinuation of alveolar bone cortical (arrow) surrounded by sclerotic trabecular bone involving the posterior region of the right mandible. Hyperdense masses were also observed in the right and left maxilla. No cortical bone destruction or expansion was noticed (Fig. 3). In the light of clinical and imaging findings we established the diagnosis of chronic osteomyelitis in association with FOD.
DISCUSSION

Usually, FOD is an asymptomatic benign condition and no treatment is required (Sadda & Phelan). However, secondary infection may occur with pain, mucosal ulceration, exposure of bone, fistula and swelling (Bencharit et al., 2003). In this article we present a case of chronic osteomyelitis in an edentulous patient conservatively treated with antibiotics and surgical curettage.

The development of low-grade osteomyelitis in edentulous areas is pointed by Melrose et al. as the principal complication of FOD. It is believed that FOD avascular nature contribute to the susceptibility to infection (Sadda & Phelan). Alsufyani & Lam investigated 118 cases of bone dysplasia treated between 1990 and 2007. Twenty five (21.2 %) were diagnosed with FOD and 93 cases (78.8 %) were periapical osseous dysplasia. Osteomyelitis was present in 6 (24 %) of the patients with FOD and in 7 (8.8 %) of the patients with periapical osseous dysplasia (Alsufyani & Lam). This higher incidence of osteomyelitis associated to FOD is probably due to the fact that FOD produces larger and more diffuse areas of sclerotic mineral tissue when compared to periapical bone dysplasia.

Some radiographic and clinic aspects should be considered to perform the differential diagnosis of FOD and other conditions like periapical and focal osseous dysplasias, fibrous dysplasia, familial gigantiform cementoma, Paget’s disease, chronic diffuse sclerosing osteomyelitis and osteosarcoma. In the present case due to the diffuse presentation of the lesions and the presence of osteomyelitis, we emphasize the importance of FOD differential diagnosis with FGC, Paget’s disease and chronic diffuse sclerosing osteomyelitis (CDSO) (Schneider & Mesa, 1990; Sarmento et al., 2013). FGC is characterized by an autosomal dominant inheritance with variable expression with a history of familial involvement. Usually FGC acomits young Caucasian patients and presents jaw bone expansion, Paget’s disease and FOD present similar radiographic features, however Paget’s disease is characterized by entire mandible involvement, loss of lamina dura, deformities of multiple bones and biochemical serum changes, such as elevated alkaline phosphate levels (Kutluay Köklü et al.; Sarmento et al.). In CDSO usually the lesion is present in one site of the mandible and the lesions are first an inflammatory process presenting swelling and tenderness of the mandible. Radiographically, CDSO appears as a single poorly delineated opaque segment of the mandible and the lesion can involve from the alveolus to the inferior margin and may extend into the ramus. Furthermore, middle-aged black women are not particular susceptible to CDSO (Schneider & Mesa).

It has been demonstrated that osteomyelitis of the jaws are usually associated to mixed anaerobic infections caused by oral microorganisms.
(Gaetti-Jardim Júnior et al., 2010). In the present article, the atrophy of the alveolar ridge associated with local trauma of the prosthesis and secondary infection by the oral microorganisms are probably the main etiopathogenic factors of the osteomyelitis lesion.

Clindamycin is a lincosamide antibiotic that acts inhibiting bacterial protein synthesis (Smieja, 1998). It has excellent oral bioavailability, bone penetration and is generally active against most anaerobes. Hence, it represents an interesting choice for the antibiotic treatment of osteomyelitis of the jaw (Fraimow, 2009). Nevertheless, due to the poor tissue diffusion in FOD lesions, complicating osteomyelitis conservative treatment only with antibiotics may be insufficient, thus surgical debridement, curettage, sequestrectomy or, in more severe cases, resection may also be required (Das et al., 2013; Kutluay Köklü et al.; Sadda & Phelan). In the case presented, osteomyelitis did not involve the entire FOD lesion. As the FOD lesion affected an extensive area of the mandible, complete resection would not be recommended, since it would lead to a big defect and morbidity. So, we opted for the conservative treatment with clindamycin, surgical exposure of the necrotic bone with curettage and follow-up.

The risk of delayed alveoli healing and bone sequestration after dental extraction in patients with FOD have been reported (More et al., 2012). Therefore, in asymptomatic patients with FOD, in order to prevent secondary infection and osteomyelitis, periodical follow-up with prophylactic management for periodontal diseases and care to avoid the need for extractions should be carried (Sadda & Phelan). Pulpar necrosis should also be prevented as the endodontic treatment may push bacteria to the periapex starting an osteomyelitis (Schneider et al., 1987). Dental rehabilitation is also challenging in patients with FOD. In edentulous patients, while the normal alveolar bone undergoes atrophy the sclerotic masses remain and the local trauma of the tissue-borne dentures can lead to secondary infection of FOD lesions. Additionally, due to the compromised vascularization and plasticity of the cancellous bone, implants osseointegration might be impaired (Sadda & Phelan; Oliveira et al., 2014). Orthodontic treatment may also impair in complications and should be carried with special attention with oral hygiene (Minhas et al., 2008).


RESUMEN: La displasia ósea florida (DOF) es una forma de displasia ósea donde el hueso trabecular normal es sustituido por tejido conectivo fibroso y tejido mineralizado amorfo. En general, las lesiones son de origen asintomático y los pacientes deben ser seguidos con el examen clínico. Sin embargo, debido a la pobre vascularización de la lesión y al trauma local, pueden producirse lesiones secundarias y osteomielitis. Los pacientes pueden presentarse con dolor, ulceración mucosa, lesión de exposición en la cavidad oral, fístula y edema. En estos casos, el correcto diagnóstico y tratamiento de la lesión es decisivo para reestablecer la salud y la calidad de la vida. El objetivo de este artículo consistió en presentar un caso de complicación secundaria crónica de osteomielitis tratados con tratamiento conservador. El paciente, negrero, de 68 años de edad, consulta por "hinchazón de las encías" que se presentó durante once años, con episodios de dolor. En el examen intraoral, se observó una púpula con supuración en la mucosa alveolar en el lado derecho de la mandíbula. La radiografía panorámica y CBCT mostraron una lesión mixta rodeadad de hueso esclerótico. El paciente fue diagnosticado con osteomielitis crónica en asociación con DOF y fue tratado con tratamiento antibiótico junto con curetaje quirúrgico. La incidencia, la etiopatología, el diagnóstico diferencial, el tratamiento y la prevención de la osteomielitis secundaria asociada con DOF se discuten a la luz de la literatura. Esta información puede ayudar a los dentistas a elegir las mejores opciones de tratamiento para casos similares.

PALABRAS CLAVE: Displasia cemento-ósea florida, Osteomielitis.

REFERENCES


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