

Florid Osseous Dysplasia complicated by a Cutaneous Sinus Tract: A Rare Case Report

Belmehdi Akram¹, El Harti Karima², El Wady Wafaa³

1-Resident in the department of oral surgery, Dental Center of Treatment and Diagnosis (Ibn Sina Hospital), Rabat, Morocco. 2-Professor of oral surgery, Faculty of Dentistry of Rabat, Mohammed V University, Morocco. 3-Professor and head of the oral surgery service, Faculty of Dentistry of Rabat, Mohammed V University, Morocco.

Correspondence to:
Dr. Belmehdi Akram, 1069, El Menzah, CYM,
Rabat, Morocco.
Contact Us: www.ijohmr.com

ABSTRACT

Florid osseous dysplasia (FOD) is an uncommon pathology of jaws which is the normal architecture of the bone and connective tissue are replaced by cemento-osseous tissue affecting the jaw bones. This lesion is most commonly seen in middle aged black women, and it's usually asymptomatic and no treatment is required. However, other symptomatic cases are almost associated with a secondary infection and its treatment can be complicated. This paper presents a rare case of FOD that was complicated by a cutaneous sinus tract. Radiological and clinical features of OD and its management will be then discussed on the basis of recent literature.

KEYWORDS: Florid osseous dysplasia, fibro-osseous lesions, cutaneous sinus tract, Complication

INTRODUCTION

Fibro-osseous lesions are a variant of benign bone disorder where normal bone is replaced by connective tissue containing of cementum or osseous tissue. The term osseous dysplasia (OD) was adopted by the World Health Organization (WHO) in 2005¹ to describe these idiopathic processes located in the periapical region tooth bearing jaw areas and characterized by fibrous tissue and metaplastic bone appearance instead normal bone.²

The WHO has also indicated a preference for the term "osseous dysplasia" but has acknowledged cemento-osseous dysplasia as a synonym³, because cementum cannot really be identified in histologic specimens nor can it be identified with any certainty on radiographic images. Various references have in the past also reported that a histologic identification of cementum is difficult to establish or confirm. The mineralized masses are more comfortably referred to as amorphous woven bone patterns.⁴

The first comprehensive clinical, radiographic and histopathologic study by Stafne dates back to 1933 where they described OD in association with apices of the teeth.² ODs are subdivided into four subgroups: (1) Periapical: affecting the periapical tissues of one or more teeth; (2) Florid: lobulated masses of dense, highly mineralized, virtually acellular cemento-osseous tissue typically occurring in several parts of the jaws and (3) Focal lesions: which share some of the features of periapical cemental dysplasia and/or florid OD, but lack their characteristic clinicopathological patterns; and 4) Familial gigantiform cementoma which is the rarest pattern of OD.²

The process may be totally asymptomatic and in such cases, the lesion is detected when radiographs are taken

for some other purposes. When a secondary infection occurs and the lesion becomes symptomatic, the management is problematic because FOD lesions cannot be treated only by the administration of antibiotics.

We report a case of FOD that was complicated by a cutaneous sinus tract. Radiological and clinical features of OD and its management will be then discussed on the basis of recent literature.

CASE REPORT

A 50-year-old white woman was referred to our department of oral surgery, because an onset of a painless swelling and persistence of a cutaneous sinus tract in the left submandibular area. Symptoms started two years ago with episodes of swelling, suppuration and regression but always with persistence of cutaneous sinus tract. The patient was healthy with no systemic symptoms.

The extraoral examination showed a facial asymmetry due to the swelling of the left mandibular area, and showed also a cutaneous sinus tract in the left submandibular area with a bloody purulent exudate that is surrounded by a quiet sclerotic skin and the examination of the lymph nodes was negative (figure 1).

The intraoral examination revealed a partially edentulous mandible and maxilla with a poor oral hygiene and persistence of the following teeth: 17, 15, 14, 43, 44, 45 and the roots of 13 and 23. The vestibule area of 37 and 38 presented an expansion coated with normal mucosa.

Orthopantomogram showed multiple well-defined sclerotic masses with a radiolucent border in both rights and left the molar region of the mandible just below the summit of the alveolar crest. These sclerotic masses were surrounded by a thin radiolucent border in the left

How to cite this article:

Belmehdi A, El Harti K, El Wady W. Florid Osseous Dysplasia complicated by a Cutaneous Sinus Tract: A Rare Case Report. *Int J Oral Health Med Res* 2017;3(5):59-62.



Figure 1: Extraoral view showing a sinus tract with a bloody exudate

quadrant of the mandible and appeared to be unattached to the root apices of the 43, 44 and 45. The retained diagnosis was of florid osseous dysplasia (figure 2).

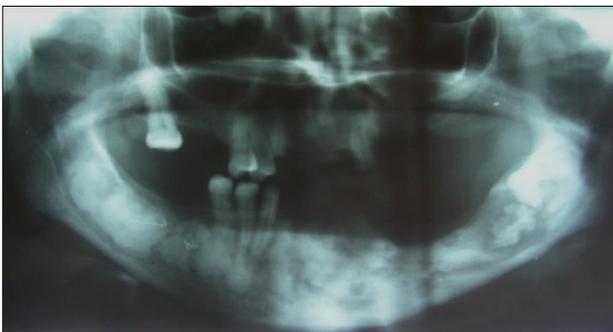


Figure 2: Orthopantomogram showed multiple well-defined sclerotic masses with radiolucent border in both right and left molar region of the mandible

The patient was then treated with antibiotics to eradicate the infection. An excision of the hard mass was conducted under local anesthesia (figure 3).



Figure 3: Surgical treatment: after incision and flap elevation, the hard mass is covered by a fibrous tissue

The anatomopathological exam was compatible with the diagnosis of FOD and the examination revealed the presence of an amorphous and dense cement bone like structures in fibrous stroma consisting of fibroblastic

cells. A follow up was made only 4 months after surgical treatment (figure 4).

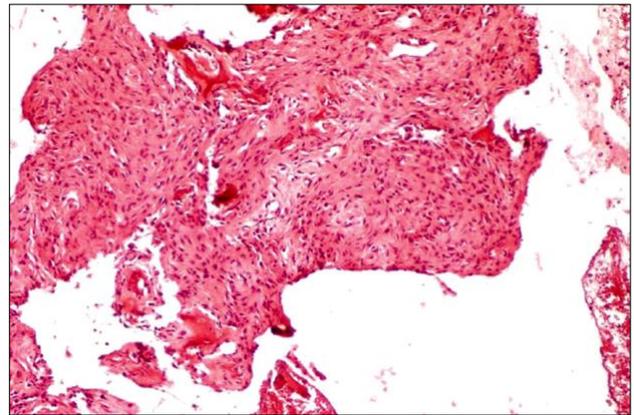


Figure 4: Presence of an amorphous, acellular eosinophilic matrix resembling cement, within a fibrous stroma. (H&E stain, $\times 10$)

DISCUSSION

Osseous dysplasias of the jaws are the most common of the fibro-osseous lesions. These lesions are more commonly referred to as cemental dysplasias, cemento-osseous dysplasias or cementomas. However, recent publications suggest that references to the term cementum should be discontinued.⁵ The rationale is that cementum cannot really be identified in histologic specimens nor can it be identified with any certainty on radiographic images. Various references have also reported in the past that a histologic identification of cementum is difficult to establish or confirm. The mineralized masses are more comfortably referred to as amorphous woven bone patterns.⁴

In the current classification of bone-related lesions, released in 2005 by the World Health Organization¹, ODs are idiopathic processes located in the periapical region of tooth bearing jaw areas and characterized by fibrous tissue and metaplastic bone appearance instead normal bone.^{2,6} These encompass several entities with the sub-classification based on differences in location and number of lesions. Periapical (Pa-OD) and focal osseous dysplasias (Fo-OD) can be lumped together as it is now thought that they are both essentially the same reactive lesion and together they constitute the most common fibro-osseous lesions of the jaws.⁷ Two other types of OD are more extensive: FOD and familial gigantiform cementoma, occurring bilaterally in the mandible or even involving all 4 jaw quadrants.¹

Regarding FOD, there are well-known sex predilections with a striking predominance of females (89%) and also racial differences exist with 64% of cases seen in black patients with a comparable finding in other studies, our case report present an unusual apparition in a white woman. Mean age at presentation ranges from 37.8 to 42.5 years with the largest proportion of cases diagnosed between the 3rd and 5th decades.⁷

The pathogenesis of the condition remains unclear. The proliferation of the fibroblastic mesenchymal stem cells

in the apical periodontal ligament (PDL) which are cementoblastic precursor stem cells, or the occurrence from the remnants of the cementum left after tooth extraction, are the most accepted theories by some authors.^{8,9} Waldron¹⁰ proposed that reactive or dysplastic changes in PDL might be the cause. Some authors attribute to the trauma from a deep bite or heavy bite causing attrition of the teeth that may activate and cause proliferation of the fibroblasts in PDL causing FOD.^{9,10} However, we tend to disagree with the proposition of deep bite/ occlusal trauma as contributing factors leading to FOD as these conditions are uncommonly seen in males whose bite force is greater than the females.⁹

Kawai and al^{11,12} showed a suggestion that these lesions are not of periodontal ligament origin but of medullary bone origin, because all the studied teeth related to the lesion had normal periodontal ligament spaces

This disorder may also have a genetic component. The familial form is reported to be an autosomal dominant trait with variable expression. It is also believed that the familial form is clinically and pathologically different from the nonfamilial form.¹²

FOD is most often asymptomatic,^{2,6,7,9} Complicated symptoms like dull pain or drainage are always associated with exposure of sclerotic calcified masses in the oral cavity. This may occur as a result of progressive alveolar atrophy under a denture or after the extraction of teeth in the affected area.⁶ Half are discovered as incidental findings; 48% first present with pain, 31% with swelling, and 30% with a discharge or a fistula.^{9,12,14,15} In our case, the main reason for consultation was the appearance of the cutaneous fistula.

The absence of symptoms typically leads to this lesion being identified through incidental findings on routine radiographic evaluation or a radiograph taken for some other reason.^{6,16}

Radiographically, because these lesions develop with time, there is a variable range of presentation dependent on the chronicity of the pattern.⁴ The lesion varies depending on the stage: predominantly radiolucent, predominantly radiodense or mixed. The location of the lesion is usually confined to the periapical alveolar area, although an aggressive lesion may expand more inferiorly and superiorly. In addition, the lesions are located in two or more quadrants. The lesion opacifies progressively as it becomes more mature.⁶

The classic appearance, as our case reported, includes diffuse, lobular, irregular-shaped radiopacities throughout the alveolar process of maxilla and mandible. Lesion size's can vary from less than 1cm to 10cm. In the mandible, a FOD lesion does not reach the inferior border; they do not occur in the rami, and they are found superior to the inferior alveolar nerve.^{12,15,17}

Diagnosis is readily achieved by conventional radiography. Although there is little need for CT in the uncomplicated case, CT (both helical and cone-beam) has revealed some more detail not appreciated by

conventional radiography, such as the central positioning of the osseous dysplastic tissue within the lesion.¹⁵

Some other lesions with similar features of FOD, and the differential diagnosis must be made with Paget's disease of bone, Gardner's syndrome, chronic diffuse osteomyelitis and cement-ossifying fibroma.^{8,9,17} Cemento-ossifying fibroma exhibits more buccolingual expansion than the FOD whereas chronic diffuse sclerosing osteomyelitis is generally unilateral with soft tissue swelling, fever and lymphadenopathy affecting primarily mandible with cyclic episodes of pain and is not always limited to the tooth-bearing areas.^{8,9} Sometimes it may be the complication of FOD.⁸

The microscopic appearances of FOD are similar with other fibro-osseous lesions, can vary depending on the maturity of the lesion and proportion of mineralized to unmineralized tissue. Again, the basic pathological process is that of replacement of bone with cellular fibroblastic tissue within which there is deposition of bony trabeculae and/or rounded cementum-like masses.⁷ Occasional spherical "cementicle" calcifications may also be seen. In more mature lesions, there may be woven, and lamellar bone and cementum-like tissue coalesce into fused sclerotic masses of globular basophilic calcifications. There may be a prominent reversal and resting lines giving a pagetoid appearance.^{12,17,18}

A simple bone cyst can be found in association with benign FOD, some reports have described an association between solitary bone cysts and fibro-osseous lesions including fibrous dysplasia and osseous dysplasias. FOD with concomitant simple bone cysts is not common.¹²⁻¹⁹

These lesions are generally considered self-limiting. However, because of their propensity to expand and possibly develop simple bone cysts, osteoplasty and biopsy are often indicated.⁴ Treatment is required when secondary infection complicated the pattern. In a edentulous area, minor irritation from a denture may cause serious consequences.¹²⁻²⁰

Surgical intervention is required when conservative treatment, such as using antibiotic therapy or washing the mouth, fails to resolve the infection. Surgical treatment through the removal of the sclerotic masses and sequestrum is effective for these cases.^{4,6,20} However, whenever surgical treatment is performed, the lack of vascularity of the lesion should be considered.⁶

We also performed the removal of sclerotic masses in mandible with the patient under local anesthesia and achieved good wound healing. Furthermore, a few aggressive forms of FOD have been reported; they required mandibular resection, followed by reconstruction of the resulting defect using vascularized graft.⁶⁻²⁰

In some cases, the debridement of necrotic infected tissue may be extensive, creating surgical defects that lead to challenging surgical and prosthodontic management. Bone grafting and implants have been successful in challenging cases.⁴⁻²⁰

The patient should be regularly followed up and recalled for examinations with prophylaxis and support of good oral hygiene care to control periodontal disease and prevent tooth loss.⁶⁻¹³

CONCLUSION

FOD represents a rare entity of jaw bones and diagnosed principally by its clinical, radiological and histopathological features.

There is no clear consensus on the management of FOD, but it is recognized that once the diagnosis is made, clinical and radiological follow-up is required as well as prevention of infectious complications, both endodontic and periodontal.

As the condition remains asymptomatic, no surgical treatment is required. However, a secondary infection may occur and its treatment can be difficult and complicated and may not be satisfactory. A long-term follow-up is carried out to assess the progress of the condition.

REFERENCES

- Barnes L, Eveson JW, Reichart P, Sidransky D. World Health Organization Classification of Tumours: Pathology and Genetics of Head and Neck Tumours. Lyon:IARC Press; 2005.
- Singh, P., Shergil, A., Solomon, M., & Carnelio, S. (2016). Osseous dysplasia: An aggressive swelling at an uncommon site. *Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology*, 28(3), 264-266.
- Sinanoglu A, Kan B, Gunhan O, Muezzinoglu B. Case of osseous dysplasia with an unusual concentric,circular, radiological appearance. *British Journal of Oral and Maxillofacial Surgery* 54 (2016) e7–e9
- Abramovitch K, Rice DD. Benign fibro-osseous lesions of the jaws. *Dent Clin North Am*. 2016 Jan;60(1):167-93.
- White SC, Pharoah MJ. *Oral radiology: principles and interpretation*. 7th edition. St. Louis (MO): Elsevier; 2014.
- Doi R, Fujii N, Okamoto A, Okamoto H, Kidani K, Takubo K, Kodani I, Ryoike K. Florid osseous dysplasia: A case report and literature review. *Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology* 26 (2014) 535–539.
- Hall J. Fibro-osseous lesions of the head and neck. *Diagnostic Histopathology*, Volume 18, Issue 4, April 2012, Pages 149-158
- Mehta RV, Khan S. Floridcement-osseousdysplasia: A case report. *Indian Dent Res Rev* 2011;3:22-3.
- Das BK, Das SN, Gupta A, Nayak S. Florid cemento-osseous dysplasia. *J Oral Maxillofac Pathol* 2013;17:150.
- Waldron CA. Fibro-osseous lesions of the jaws. *J Oral Maxillofac Surg* 1985;43:249-62.
- Kawai, T.; Hiranuma, H.; Kishino, M.; Jikko, A. & Sakuda, M. Cemento-osseous dysplasia of the jaws in 54 Japanese patients: A radiographic study. *Oral Surg. Oral Med. Oral Pathol. Oral Radiol. Endod.* 87:107-14, 1999.
- Benjelloun L, El Harti K, El Wady W. Florid osseous dysplasia: Report of two cases and a review of the literature. *Int J Odontostomat*, 5(3):257-266, 2011.
- De Sarmetro DJ, Brito Metro BV, de Medeiros AM, da Silveira EJ. Severe florid cemento-osseous dysplasia: a case report treated conservatively and literature review. *Oral Maxillofac Surg* 2013;17:43–6.
- Schajowicz FM. Histological typing of bone tumours. In: *International histological classification of tumours*. 2nd ed. London: Springer-Verlag; 1993. p. 39e40.
- MacDonald DS. Maxillofacial fibro-osseous lesions. *Clinical Radiology* 70 (2015) 25-36.
- Dagistan S, Tozoglu U, Goregen M, Cakur B. Florid cemento-osseous dysplasia:a case report. *Med Oral Patol Oral Cir Bucal* 2007;12:E348–50.
- Marx, R. E. & Stern, D. *Oral and maxillofacial pathology: a rationale for diagnosis and treatment*. 2nd ed. Illinois, Quintessence Publishing, 2012.
- Speight PM, Carlos R. Maxillofacial fibro-osseous lesions. *Curr Diag Pathol*, 12:1-10, 2006.
- Rao KA, Shetty SR, Babu SG, Castelino RL. Co-Occurrence of Florid Cemento-Osseous Dysplasia and Simple Bone Cyst: a Case Report. *J Oral Maxillofac Res* 2011;2(3):e5
- Bencharit S, Schardt-Sacco D, Zuniga JR, et al. Surgical and prosthodontic rehabilitation for a patient with aggressive florid cemento-osseous dysplasia: a clinical report. *J Prosthet Dent* 2003;90(3):220–4.
- Lopes MA, Kim HS, Mariano FV, Correa MB, Rabelo NT, Vargas PA. Clinico-pathologic conference: case 1. High-grade osteosarcoma (OS) and florid cemento-osseous dysplasia (FCOD). *Head Neck Pathol* 2010;4:329–33.

Source of Support: Nil
Conflict of Interest: Nil