Florid Cemento-osseous Dysplasia Associated with Secondary Infection - A Case Report

Abrar Khalid Demyati

Department of Oral and Maxillofacial Surgery & Rehabilitation, Faculty of Dental Medicine, Umm Al-Qura University, Mecca, Saudi Arabia

Abstract

Rationale: The term cemento-osseous dysplasia (COD) refers to a bony fibro-osseous lesion, in which fibrous tissue and cementum-like tissue replace normal bone. There are three types of COD: periapical, focal and florid. The condition is usually asymptomatic and treatment is unnecessary; however, a secondary infection could occur, which requires treatment. **Patient Concern:** A 58-year-old female patient presented with symptoms in the mandibular posterior region of the right jaw for six months. **Diagnosis:** Infected florid COD (FCOD). **Treatment:** A pre-operative antibiotic, followed by extraction of non-restorable teeth, debridement of the infected tissue and necrotic bone removal. **Outcome:** The patient was followed for one year, during which all previously reported signs and symptoms were resolved. **Take-away Lessons:** Early lesion detection is essential. Treatment depends on the presence or absence of clinical and radiographic manifestations. The current case was treated surgically to minimise complications.

Keywords: Cemento-osseous dysplasia, florid, focal, infection, periapical

Introduction

Cemento-osseous dysplasia (COD) is a non-neoplastic benign fibro-osseous lesion (FOL) that frequently occurs in the jaw in the tooth-bearing area or edentulous alveolar process.[1] Depending on the lesion's presentation and location, COD can be classified as periapical, focal or florid.[2] The extensive form is florid COD (FCOD), which appears in more than one jaw quadrant.[1] In COD, the normal bone tissue is continuously replaced by fibrous and calcified tissue, resembling the bone and cementum.[3] COD appears commonly in the mandible above the mandibular canal in the alveolar bone and the adjacent teeth remain vital. It is recognised as an odontogenic tumour and originates from the periodontal ligament.[2] COD is common and is more likely to present in women between 30 and 50 years of age.[4] It occurs in multiple racial groups, including Africans, followed by the Asians and a few cases have been reported in Caucasians.[4,5] The reasons for the gender and racial tendencies are undetermined.[4]

COD is usually asymptomatic and the covering gingiva remains healthy.[4] It is diagnosed incidentally during routine dental radiographic examinations.[1] On the radiograph, the early stage of the lesion appears radiolucent, but as it matures, it shows a mixed appearance of radiolucency and radiopacity.[4] Later, it shows more radiopacity as diffuse sclerosis with a radiolucent halo margin.[4] However, inflammatory signs may be present if infection of the lesion occurs.[4] These signs include but are not limited to local swelling, discharge, redness and pain.[4] Treatment planning is influenced by the presence or absence of clinical and radiological manifestations.[1] This study reported a case of FCOD including the relevant clinical and histologic features, radiologic characteristics, differential diagnoses and management options.

Case Report

A 58-year-old female patient was referred to the Department of Oral and Maxillofacial Surgery in November 2020 for

Access this article online

Quick Response Code:

Website: https://journals.lww.com/aoms

DOI: 10.4103/ams.ams_49_23

Address for correspondence: Dr. Abrar Khalid Demyati, Department of Oral and Maxillofacial Surgery & Rehabilitation, Faculty of Dental Medicine, Umm Al-Qura University, Mecca 2373, Saudi Arabia.
E-mail: akdemyati@uqu.edu.sa

Received: 12-03-2023 **Last Revised:** 26-06-2023
Accepted: 31-07-2023 **Published:** 26-09-2023

How to cite this article: Demyati AK. Florid cemento-osseous dysplasia associated with secondary infection - A case report. Ann Maxillofac Surg 2023;13:232-5.

treatment of an infection related to the right mandibular area [Figure 1]. Her chief complaint was throbbing intermittent pain and buccal swelling in the right mandibular posterior region for the past months. Furthermore, she reported bad taste and abnormal secretion three weeks ago.

The patient's medical history revealed findings of controlled type 2 diabetes mellitus and hypertension. She was managing her diabetes through diet and prescribed oral hypoglycaemic medications. In addition, she was taking calcium channel blockers and long-acting angiotensin-converting enzyme inhibitors to control hypertension. In her dental history, the patient reported multiple teeth extractions. A clinical evaluation revealed poor oral hygiene, pus and bleeding related to grossly decayed right lower first and second molar teeth. In addition, she has multiple decayed teeth, root stumps, missing teeth, periodontal disease, and her bite was shown to be collapsed. The patient brought an old panoramic radiograph before starting any treatment, which depicted a calcified radio-opaque mass in the apical region of the remaining root of the right mandibular first molar (#46). In addition, a smaller radio-opaque area was seen in the edentulous area corresponding to the left mandibular second molar (#37) and other multiple grossly decayed teeth [Figure 2]. A new panoramic radiograph was obtained when the patient was referred to the Department of Oral and Maxillofacial Surgery [Figure 3]. For further pre-operative evaluation, cone-beam computed tomography was performed [Figure 4a and b].

Provisional diagnoses of a FOL, focal sclerosing osteomyelitis and periapical abscess was suggested. The patient was scheduled for surgery because of the presence of symptoms. A pre-operative antibiotic (amoxicillin/clavulanate) was given to the patient to treat the infection and minimise adverse effects before the surgical appointment for the debridement. Under local anaesthesia, the surgical procedure included raising a full-thickness mucoperiosteal flap to provide surgical access for extraction and debridement. Both soft-and hard-tissue biopsies were taken and sent for histopathological evaluation. The histopathological report confirmed the clinical and radiographic diagnosis of COD [Figure 5a-d]. The patient came for follow-up visits for up to one year and showed satisfactory favourable and uneventful healing. She was referred to the Department of Prosthodontics for denture fabrication [Figure 6a-d].

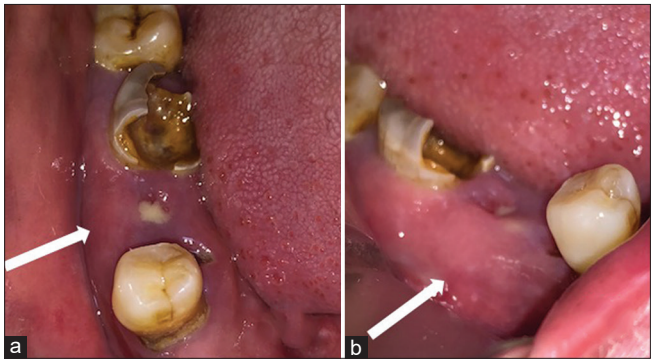

Figure 1: The right lower quadrant with grossly decayed right lower molars. (a) Occlusal view with areas of suppuration (white solid arrow); (b) Lateral view showing the swelling (white solid arrow)

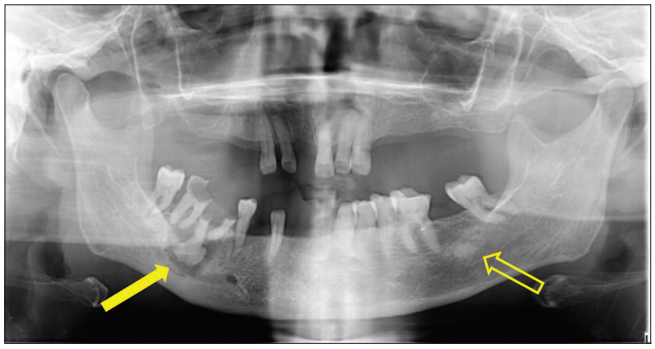

Figure 3: Pre-operative panoramic radiograph reveals a radio-opaque lesion with radiolucent margin associated with grossly decayed tooth #46 (solid arrow). Radio-opaque lesion was related to the edentulous area of tooth #37 (hollow arrow)

Discussion

COD has been described as the most common lesion of the FOL of the jaws.[5] Global literature shows that COD affects

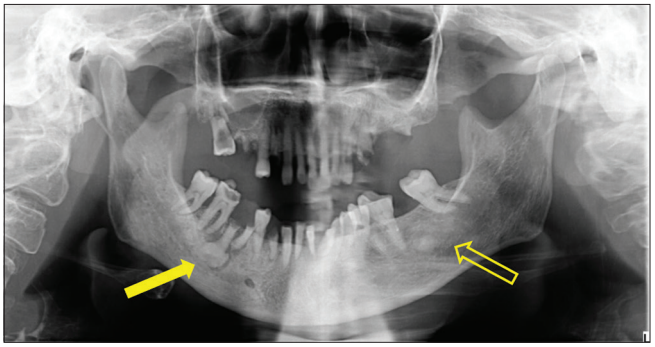

Figure 2: Pre-operative orthopantomogram. Radio-opaque lesions in the mandible localized on the periapical area of tooth #46 (solid arrow) and in the edentulous area corresponding to tooth #37 (hollow arrow). Multiple carious non-restorable teeth and periodontal disease can also be seen

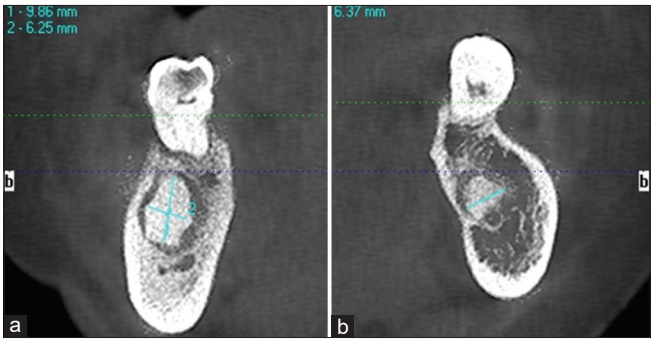

Figure 4: (a) Cone-beam computed tomography of the coronal cross section shows a well-defined radio-opacity with the greatest dimension of 9.86 mm × 6.25 mm in the right lower area. (b) A well-defined radio-opacity with the greatest dimension of 6.37 mm × 5.32 mm in the left lower area

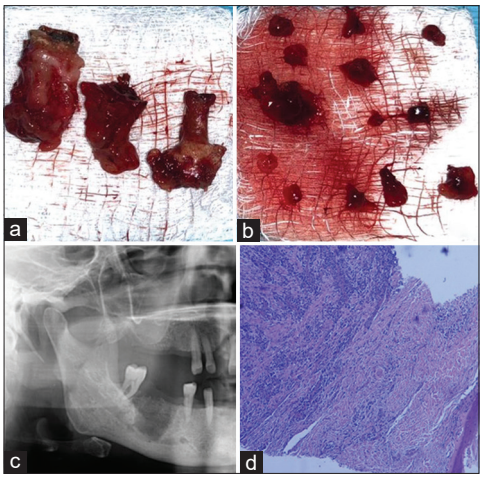

Figure 5: (a) Extracted teeth with the associated lesion. (b) Granulation tissue. (c) Immediate panoramic radiograph after surgical debridement. (d) Haematoxylin – eosin staining of a medium-power histologic cut revealing a transition of bone mass; the adjacent fibrous tissue presents an inflammatory infiltrate

30-70% of the FOL of the jaws.[5] The radiographic and histologic appearance of FCOD depends on the maturation stage of the lesion.[3] The aetiology of COD is still unknown.[2] The periodontal ligament elements and hormonal imbalance have been suggested to be causes of COD.[2,6] Our patient had carious non-restorable teeth, which could be the cause of the infection.

Clinically, although COD is generally asymptomatic, sometimes, in severe cases, there may be discomfort, dull pain, drainage or localised bone expansion.[5] Our patient had similarities to an infected COD lesion, which is associated with the development of pain, gingival swelling, suppuration, delayed wound healing after an extraction, bone sequestrum, bone exposure, focal expansion and facial deformity, with subsequent progression to necrosis.[7] Radiographically, most cases of COD are diagnosed in the mixed radiolucent-radio-opaque stage (72%).[3] The radiographic findings in our case were in agreement with the findings in the literature.[3] Similar to this case, secondary infection of the lesion can appear as periapical inflammatory radiolucency or may be associated with sequestrum into a well-defined radiolucency.[3]

Histologic analyses of early lesions show early osteoid trabeculae.[8] Over time, there is increasing additional osteoid trabeculae and fibrosis of the vascular stroma.[8] In mature lesions, cemento-osseous material coalesces with the osteoid trabeculae, which replaces the fibrous stroma.[8] When secondary infection occurs, alongside the typical characteristics of COD, the specimen shows signs of infection.[5] For the treatment, it is essential for the clinician to make an accurate diagnosis before choosing appropriate management to prevent unnecessary surgical intervention.[1]

Treatment methods depend on symptoms.[5] For example, in asymptomatic COD, no treatment should be performed because the teeth are vital and doing so will cause osteomyelitis owing to the hypovascular nature of the lesions.[5] However, long-term

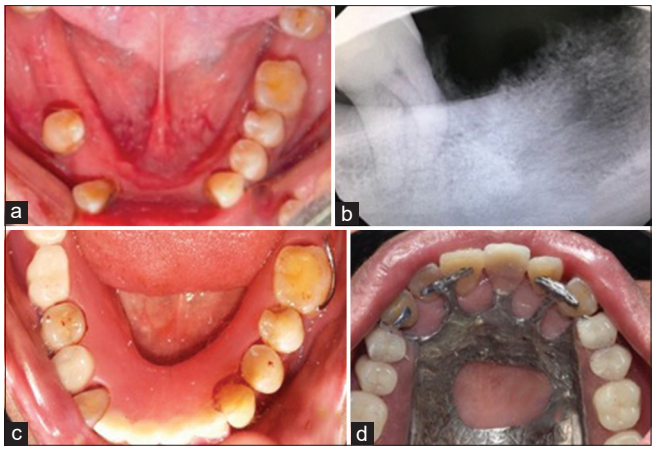

Figure 6: (a) Clinical follow-up after six months. (b) Radiographic follow-up after six months. (c) Lower interim removable partial denture. (d) Upper metallic partial denture

follow-up should be performed every two to three years.[9] If symptoms are present, despite the lack of consensus on the most appropriate therapy, curettage and removal of necrotic bone are acknowledged as an appropriate approach.[5] In addition, the literature has suggested different treatment methods for the treatment of infected COD, similar to the treatment of osteomyelitis.[10] Regarding medications, the lesion may not respond to antibiotics because of its avascular nature, which indicates the need for surgical enucleation and debridement.[9] Our patient was prescribed an antibiotic, and curettage and necrotic bone removal were the treatments of choice. As every case is unique, each patient receives a unique treatment plan based on a combination of all relevant characteristics.

Conclusion

This case of FCOD in association with infection is very unique because of the risk of osteomyelitis development and related adverse effects. Therefore, surgical intervention was recommended to obtain a favourable clinical outcome.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Nelson BL, Phillips BJ. Benign fibro-osseous lesions of the head and neck. Head Neck Pathol 2019;13:466-75.
2. El-Naggar AK, Chan JK, Grandis JR. WHO Classification of Head and Neck Tumours. Geneva, Switzerland: IARC Press, WHO Press, World

Health Organization; 2017.
3. Fokam ST, Kwedi GK, Messanga CB. Infected florid cemento-osseous dysplasia: About one clinical observation. Adv Oral Maxillofac Surg 2022;7:100298.
4. Min CK, Koh KJ, Kim KA. Recurrent symptomatic cemento-osseous dysplasia: A case report. Imaging Sci Dent 2018;48:131-7.
5. Kato CN, de Arruda JA, Mendes PA, Neiva IM, Abreu LG, Moreno A, *et al.* Infected cemento-osseous dysplasia: Analysis of 66 cases and literature review. Head Neck Pathol 2020;14:173-82.
6. Benaessa MM, Mahomed F, Ngwenya SP. A retrospective clinico-pathologic analysis of cemento-osseous dysplasia in a South African patient population. Afr Health Sci 2019;19:3154-9.
7. Kato CN, Nunes LF, Chalub LL, Etges A, Silva TA, Mesquita RA. Retrospective study of 383 cases of fibro-osseous lesions of the jaws. J Oral Maxillofac Surg 2018;76:2348-59.
8. Fenerty S, Shaw W, Verma R, Syed AB, Kuklani R, Yang J, *et al.* Florid cemento-osseous dysplasia: Review of an uncommon fibro-osseous lesion of the jaw with important clinical implications. Skeletal Radiol 2017;46:581-90.
9. Saikia J, Pachipulusu B, Govindaraju P. Florid cemento-osseous dysplasia associated with chronic suppurative osteomyelitis and multiple impacted tooth an incidental finding – A rare case report. J Family Med Prim Care 2020;9:1757-61.
10. Geurts J, Hohnen A, Vranken T, Moh P. Treatment strategies for chronic osteomyelitis in low- and middle-income countries: Systematic review. Trop Med Int Health 2017;22:1054-62.