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CASE STUDY

Mandibular Actinomyces Osteitis Associated with Florid Cemento-Osseous Dysplasia

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ABSTRACT

Introduction: **Actinomycosis is a rare chronic suppurative granulomatous infection caused by actinomyces. The Florid cemento-osseous dysplasia (FCOD) is a benign fibro-osseous lesion. We report a case of simultaneous association of these two entities with the use of PRF for the first time in this context.**

Case Report: **A 59-year-old female with no medical history was received in our clinic for painful swelling on the right mandible. On clinical examination, we noticed a facial asymmetry caused by the swelling and bone exposure with pus on the right mandibular ridge. The panoramic radiography showed right and left posterior mandibular opacities. On the cone beam reconstructions, we noted a hyperdense posterior right mandibular image surrounded by a hypodense border and a hyperdense posterior left mandibular image surrounded by a hypodense border, in contact with the apices of tooth 36. Based on the clinical, radiographic and pathological findings, the present case was diagnosed as mandibular actinomyces osteitis associated with florid cemento-osseous dysplasia. The treatment consisted of surgical excision of the lesion, filling of the defect with PRF and prescription of antibiotics.**

Conclusion: **Despite its rarity, practitioners should be aware of the possibility of the coexistence of these two entities simultaneously. Thus, they can initiate the appropriate treatment.**

Keywords: Florid cemento-osseous dysplasia, Mandibular Actinomyces osteitis, PRF.

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1. Introduction

Actinomycosis is a rare chronic suppurative granulomatous infection caused by actinomyces. About 50% of actinomycosis cases occur in the cervicofacial region. However, bone involvement is an exceptional complication, it only represents between 1% and 15% of cases of cervicofacial actinomycosis [1], [2].

Meanwhile, florid cemento-osseous dysplasia (FCOD) is a benign fibro-osseous lesion in which mature bone is replaced with woven bone in a matrix of fibrous connective tissue. It is infrequent cemento-osseous dysplasia according to the classification of head and neck tumors published by the WHO in 2017 [3], [4].

Both of the aforementioned entities are very rare. Hence, their association is even rarer. We report the second case, according to the consulted literature, presenting the simultaneous association of these two entities with the use of PRF for the first time in this context.

2. Case Report

A 59-year-old female with no medical history was received in our clinic for painful swelling on the right mandible.

On questioning, the patient stated that she had a cellulitis with cutaneous fistula in the same area which led to the extraction of the responsible tooth (tooth 47) in 2015.

The extraoral examination showed a facial asymmetry caused by the swelling and the presence of the fistula scar on the right mandible. The patient had a normal mouth

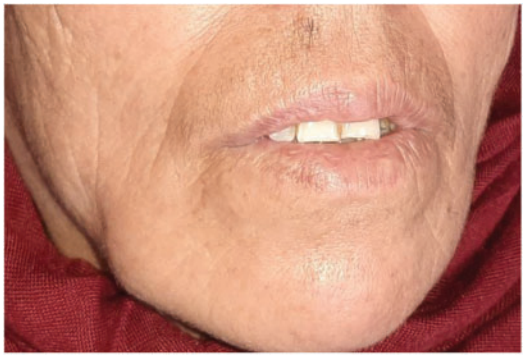

Fig. 1. Extraoral view showing a facial asymmetry caused by the swelling and the presence of the drainage scar on the right mandible.

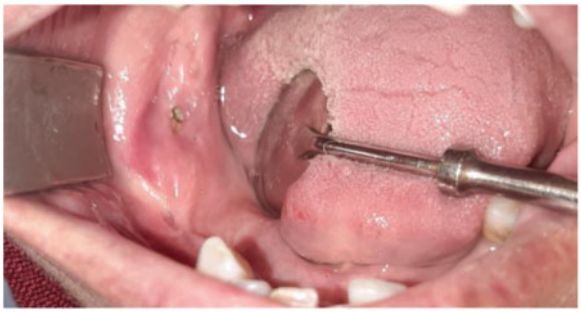

Fig. 2. Intraoral view objectifying a bone exposure on the right mandibular ridge.

opening and no sensitive problem was raised. We also noted the absence of any lymphadenopathy (Fig. 1).

The intraoral examination showed poor oral hygiene and the absence of several teeth. Moreover, it highlighted a bone exposure with pus on the right mandibular ridge (at the site of tooth 47). Palpation was painful on the buccal shelf of the right mandible (Fig. 2).

The panoramic radiography revealed a posterior right mandibular radiopacity measuring 35 mm in mesiodistal, and a radiopacity on the periapical area of tooth 36 measuring 25 mm in mesiodistal. Both images overlapped the mandibular canal. Another left mandibular radiopacity was noted measuring 8 mm in mesiodistal (Fig. 3).

We also asked for a cone beam to determine the extension of the lesion. On the reconstructions, we noted a hyperdense posterior right mandibular image surrounded by a hypodense border and a hyperdense posterior left mandibular image surrounded by a hypodense border, periapical of tooth 36. All lesions are distant from the mandibular canal (Fig. 4).

Based on both clinical and radiographic findings, our supposed diagnosis was an infected florid cemento-osseous dysplasia.

Treatment consisted of surgical excision of the lesion. Under local anesthesia, a mucoperiosteal flap was elevated, and most of the lesion was excised and conditioned for histological examination (Figs. 5 and 6).

Afterwards, we prepared the PRF membranes and venous blood was collected in 8 sterile vacutainer tubes of 4 ml capacity without anticoagulant. The vacutainer tubes were placed in a centrifugal machine at a speed of 3000 revolutions per minute (rpm) for 10 minutes. The centrifugation collected blood settled into the following layers: Red lower fraction containing red blood cells, the upper straw-colored cellular plasma, and the middle fraction containing the fibrin clot. Thereafter, the membranes were placed on the site (Fig. 7). The mucoperiosteal flap was returned and sutured (Fig. 8). Postoperative medication and instructions were advised.

The patient was seen 7 days after for control and suture removal. Clinically, the bone exposure was persisting (Fig. 9). The result of the histological examination demonstrated inflammatory lesions with actinomyces (Fig. 10). Hence, this suggested the diagnosis of actinomyces osteitis.

Based on all clinical, radiographic, and histological findings the diagnosis was a mandibular actinomyces osteitis associated with florid cemento-osseous dysplasia. Therefore, we adapted our treatment which consisted of an antibiotic prescription: amoxicillin + clavulanic acid 2 g per day until mucosal and bone healing with monitoring every two weeks.

On Clinical and radiological control of week 22, there was complete bone and mucosal healing (Figs. 11 and 12). The antibiotic prescription was stopped and we maintain the follow-up every month.

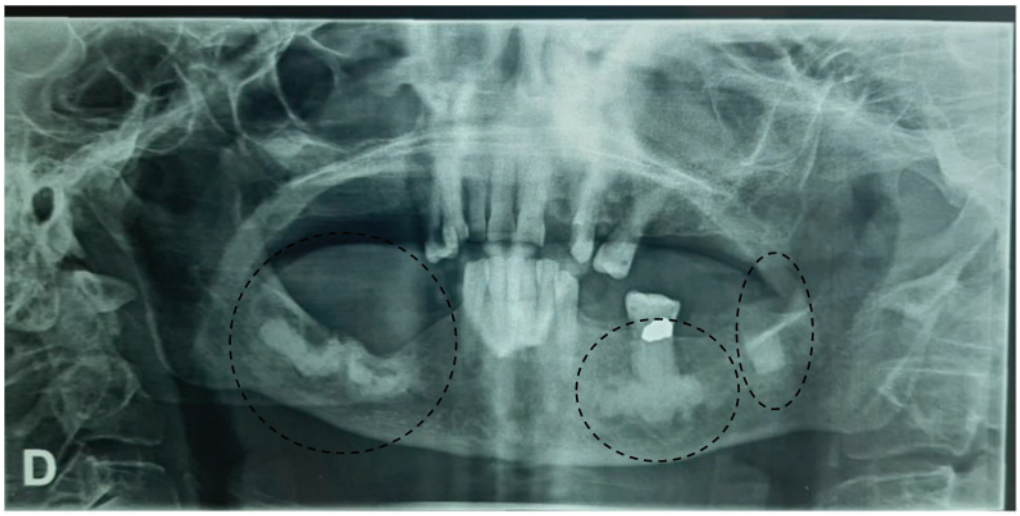

Fig. 3. The panoramic radiography revealed a posterior right mandibular radiopacity, a radiopacity on the periapical area of tooth 36 and another left mandibular radiopacity.

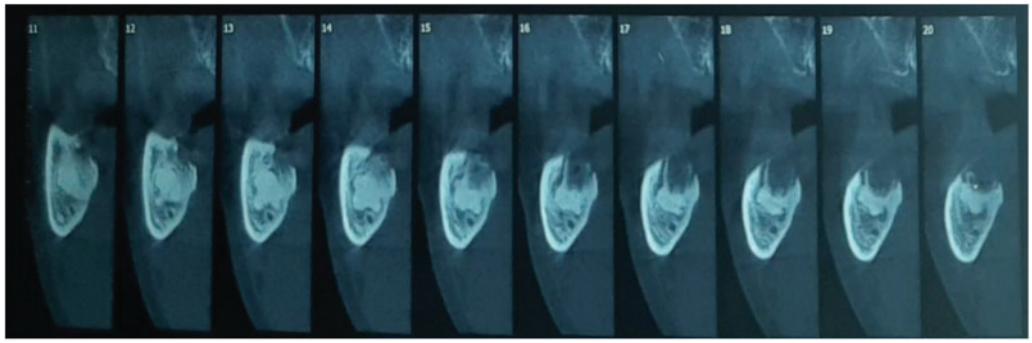

Fig. 4. The cone beam reconstructions showed: a hyperdense posterior right mandibular image surrounded by a hypodense border distant from the mandibular canal.

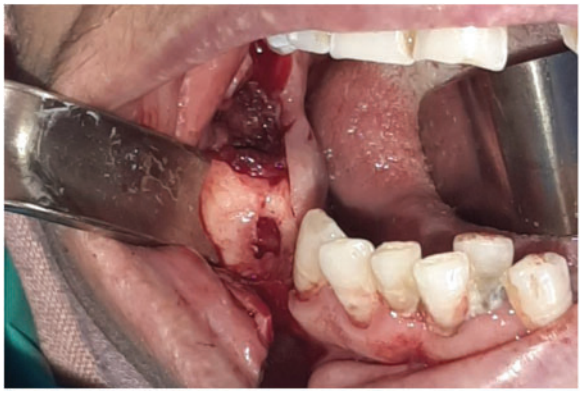

Fig. 5. Intraoral view of the elevation of the mucoperiosteal flap.

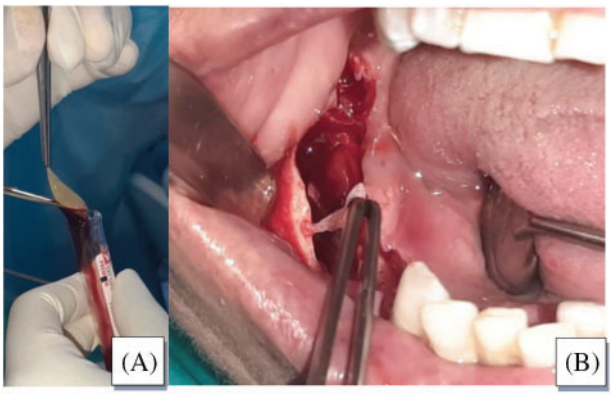

Fig. 7. A) PRF membrane and B) Intraoral view showing the filling of the defect by the PRF membranes.

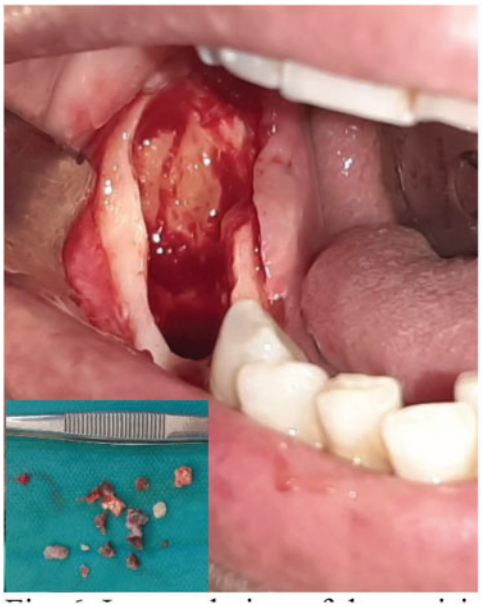

Fig. 6. Intraoral view of the excision of most of the lesion.

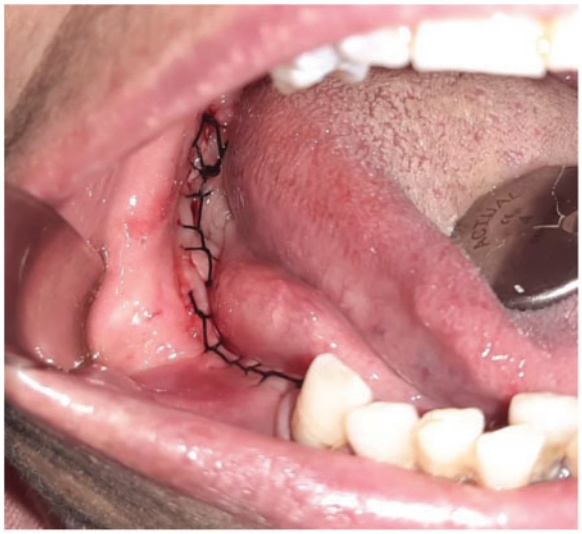

Fig. 8. Mucoperiosteal flap suture.

3. Discussion

Florid cemento-osseous dysplasia is a condition that affects middle-aged women and has a predilection for people of African and Asian descent. In our case, those attributes are valid for our patient who is an African female in her middle age. Besides, FCOD is often of an asymptomatic slow growth. They are usually diagnosed through routine radiographic examination. Still, FCOD may be characterized by pain and discharge secondary to infection which was the case for our patient [5].

The lesions manifest as multiple, lobular, well-limited intraosseous masses, which surround the root apices of vital teeth or arise in previously tooth-bearing edentulous areas. The lesions are usually bilateral and have a symmetrical appearance. This classical radiographic appearance was identified for our patient too [6].

However, it is to be mentioned that the radiographic appearance depends upon the maturation of the lesion. Over time, the radiolucency lesions transform into mixed and then opaque masses [7].

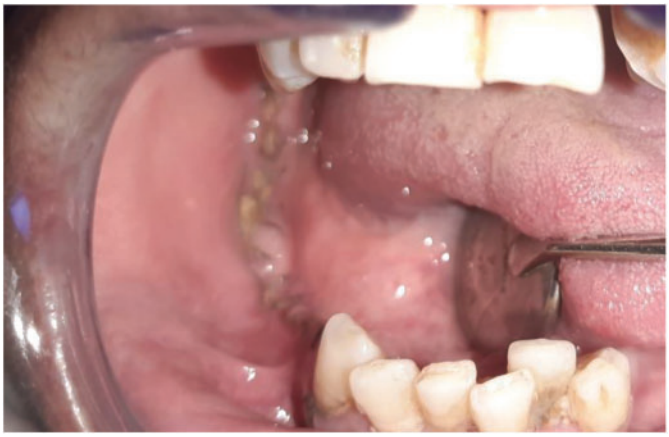

Fig. 9. Intraoral view revealing the persisting of bone exposure a week after.

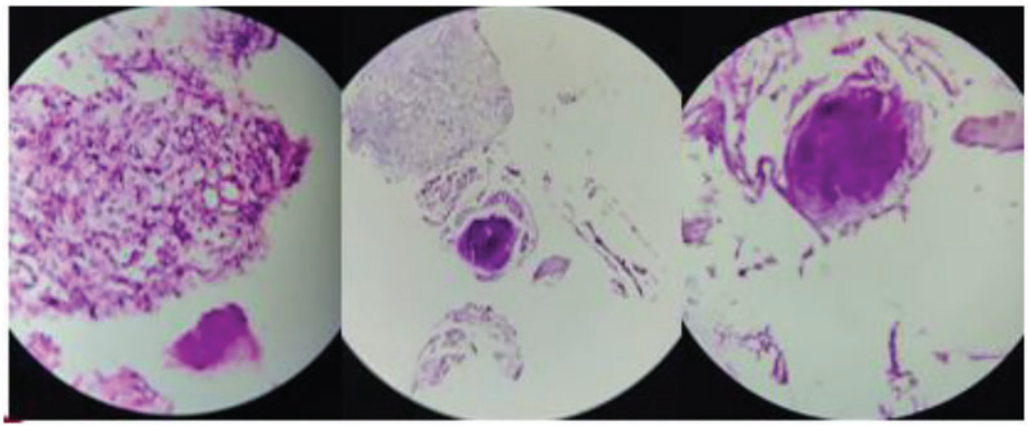

Fig. 10. The histological examination showed inflammatory lesions with actinomyces.

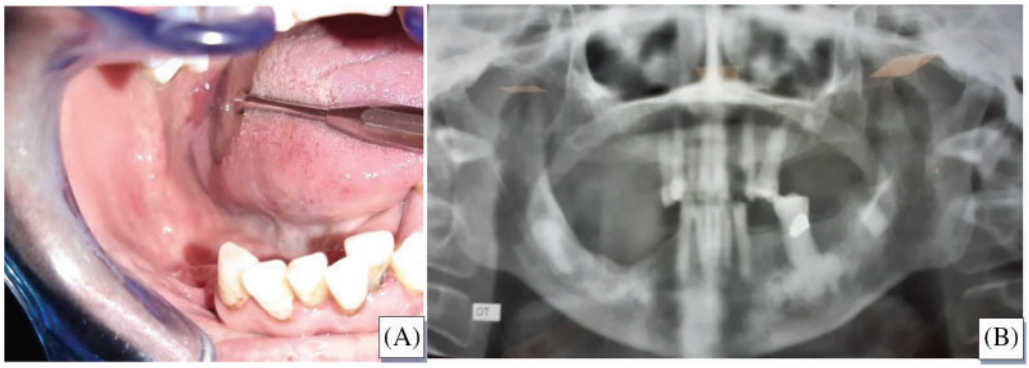

Fig. 11. A) intraoral view showing mucosal healing at week 22 and B) the panoramic radiography revealed bone healing at week 22.

FCOD is known by fibrovascular stroma with a *ginger-root* pattern of irregular curvilinear trabeculae, as well as droplets of cementum (*cementicles*) that fuse to form bosselated structures. Early-stage lesions of FCOD display fewer trabeculae and more prominent fibrovascular stroma, often with hemorrhage [8].

Furthermore, actinomycosis is a rare infection too that occurs mostly in men and adults between the ages of 20 and 60. However, our patient was a 59-year-old female [1].

The infection is caused by anaerobic gram-positive bacteria or microaerophiles, belonging to the genus Actinomyces, which are saprophytes of the oral cavity and the gastrointestinal tract [9].

The principal species identified are *Actinomymyces israelii*, *odontolyticus*, *viscosus*, *naeslundii*, or *meyeri* [10].

They become pathogenic under the influence of several local factors such as breaking of the mucosal barrier caused by dental extractions, jaw trauma, poor oral hygiene, and general factors like diabetes, immunosuppression, corticoid treatment for extended periods, alcoholism and smoking. In our case, the previous extraction of the 47 teeth and poor oral hygiene may be the etiology [11].

A clinical diagnosis of actinomycosis may be difficult. The common initial signs and symptoms of infection such as fever, sudden onset of pain, swelling, erythema and edema may be absent. Our patient only described a painful swelling with no fever. It, also, manifests as a slowly

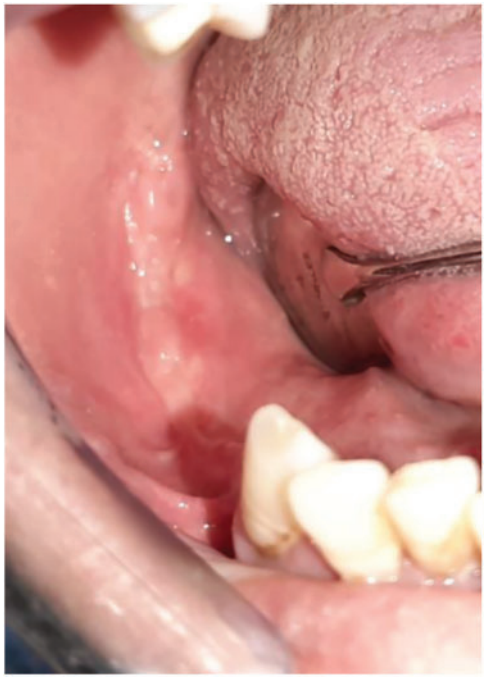

Fig. 12. Intraoral view at 28 weeks follow-up.

growing, painless, indurated mass with multiple draining sinus tracts on the skin or oral mucosa and occasionally the presence of thick, yellow exudate with characteristic sulfur granules. As for our patient, we only objectified an indurated mass with bone exposure [12].

The diagnosis of actinomycosis is confirmed by bacteriological and histopathological analysis. However, the identification of actinomycete by bacteriological analysis is often difficult due to the nature of bacteria which is hard to collect and grows slowly, and the use of antibiotics prescribed most often before the examination. For our part, we were faced with this obstacle. Hence, our definitive confirmation of the diagnosis was based on the identification of the bacteria in histological sections [13].

In the case presented here, the typical findings of FCOD were seen in conjunction with key features of actinomyces osteomyelitis including filamentous gram-positive organisms, acute inflammation, and necrotic bone [8].

The treatment combines both surgery and antibiotic therapy that consists of high doses of penicillin that, depending on the severity of the case, can be administered by intravenous penicillin G infusion in doses ranging from 3–12 million units daily over two to six weeks, followed by oral penicillin A for 6 to 12 months or oral penicillin A administration of 2–4 gr per day for a period ranging from 6 to 12 months, depending on the response of the host to the infection [14].

An alternative for allergic patients would be macrolides, clindamycin, tetracycline, erythromycin, carbapenem, imipenem, and cephalosporin. Commonly least effective antimicrobials against Actinomyces species include aminoglycosides, and metronidazole [15]. The surgical component includes debridement, curettage and sequestrectomy. As for us, we opted for both surgery and antibiotic therapy with amoxicillin + clavulanic acid 2 gr per day. It showed good results [16].

Regarding Platelet-rich fibrin (PRF), it is an autologous platelet concentrate that consists of cytokines, platelets, leukocytes, and circulating stem cells. It has been considered to be effective in bone regeneration and is mainly used for oral and maxillofacial bone regeneration [17].

It is the 2nd generation of platelet concentrates made from an anticoagulant-free blood harvest without any artificial biochemical modification [18].

It was used in our case to improve bone healing and promote its regeneration. Our radiographic controls witnessed its effectiveness and good results were obtained.

Conflict of Interest

Authors declare that they do not have any conflict of interest.

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