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Up-to Date Review And Case Report

Florid osseous dysplasia causing sensory disturbances in the area supplied by the inferior alveolar nerve

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Abstract – Introduction: Florid osseous dysplasia is a rare benign pathology, characterized by a multifocal extension, and possibly affecting the four maxillo-mandibular quartors. It is often a fortuitous discovery, and only becomes symptomatic when the lesions are voluminous, deforming mandible or maxillary, or when secondary infections occur. **Observation:** A 45-year-old African patient is referred for diagnosis of a florid osseous dysplasia. Mandibular lesions are revealed by a CBCT scan. As the patient is asymptomatic, an annual follow-up was established. Six months later, the patient shows neuropathic pain signs on the right side of the chin and the inferior lip: the inferior alveolar nerve seems to be affected. A surgical treatment is then proposed to the patient. **Discussion:** Affection of the alveolo-dental nerve due to florid osseous dysplasia has never yet been reported in literature. However, our patient showed neuropathic pain symptoms, with no sign of infection, and the CBCT revealed a great proximity of the nerve and the lesions. The surgery was a success: removing the calcified masses has allowed us to amend the neuropathic pain. We can then suppose that the bone lesions were directly responsible for our patient's pain symptoms. **Conclusion:** Florid osseous dysplasia lesions, when close to the inferior alveolar nerve, can cause a nerve illness, by compression, on its intracanal trajectory or by its foramen.

Introduction

Bone dysplasia, formerly known as cemento-osseous dysplasia, is characterized by benign bony idiopathic lesions. It originates from the alveolar ligament and is characterized by the replacement of the normal bone by fibrous tissue and metaplastic bone tissue [1,2]. It is divided into four forms depending on the location, size, and number of lesions: (i) periapical bone dysplasia involves a limited number of teeth and is located in the anterior part of the mandible or maxilla; (ii) focal bone dysplasia also involves a limited number of teeth but has a posterior localization; (iii) florid bone dysplasia involves several quadrants; (iv) the gigantiform cementoma is a single lesion that can reach a large size and primarily affects children [3,4]. These lesions are usually asymptomatic and are often discovered incidentally on panoramic X-rays. Symptom onset coincides with an increase in size or intraoral exposure of dysplastic lesions during infectious episodes. Here we discuss the case of a patient with a florid osseous dysplasia associated with neurosensory disturbance of the inferior alveolar nerve in the labiomental region.

Observation

A 45-year-old patient of African origin consulted after the discovery of bone lesions on the panoramic X-ray. She had no medical history. Her calcium/phosphate equilibrium was normal. The intraoral examination found firm and painless vestibular swelling of the mandibule with a normal mucosa (Fig. 1), and the vitality pulp testing was positive for all teeth near the bone dysplasia. The panoramic X-ray showed apical lesions of the teeth 47, 45, 44, 43, 42, 41, 31, 32, 33, 34, 35, 37 as well as decay or restorative treatments of the teeth 48, 47, 44, 37, and 38 (Fig. 2). Cone-beam computerized tomography (CBCT) has allowed a more precise characterization of the appearance, size, and anatomical ratios of the various lesions around the teeth as well as those in

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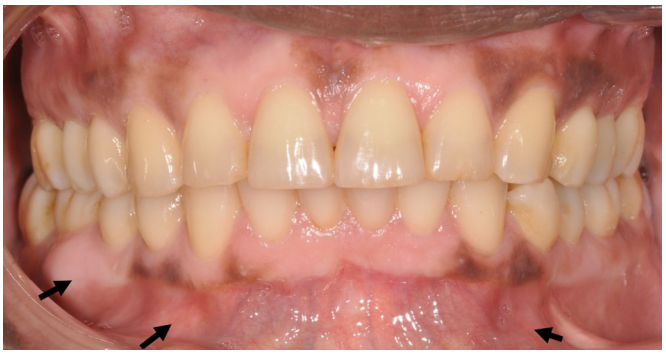

Fig. 1. Left mandibular buccal bone swelling.

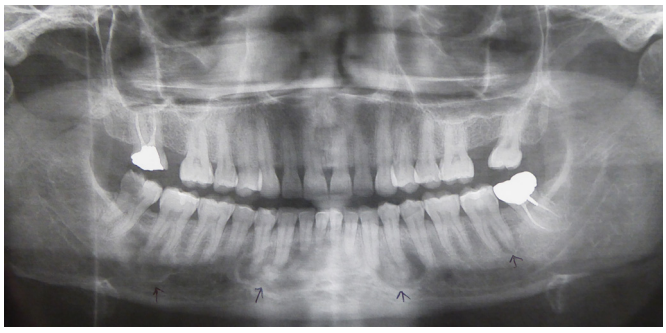

Fig. 2. Initial panoramic radiography with mixed multilobular well-defined images of the alveolar bone, respecting the basilar bone.

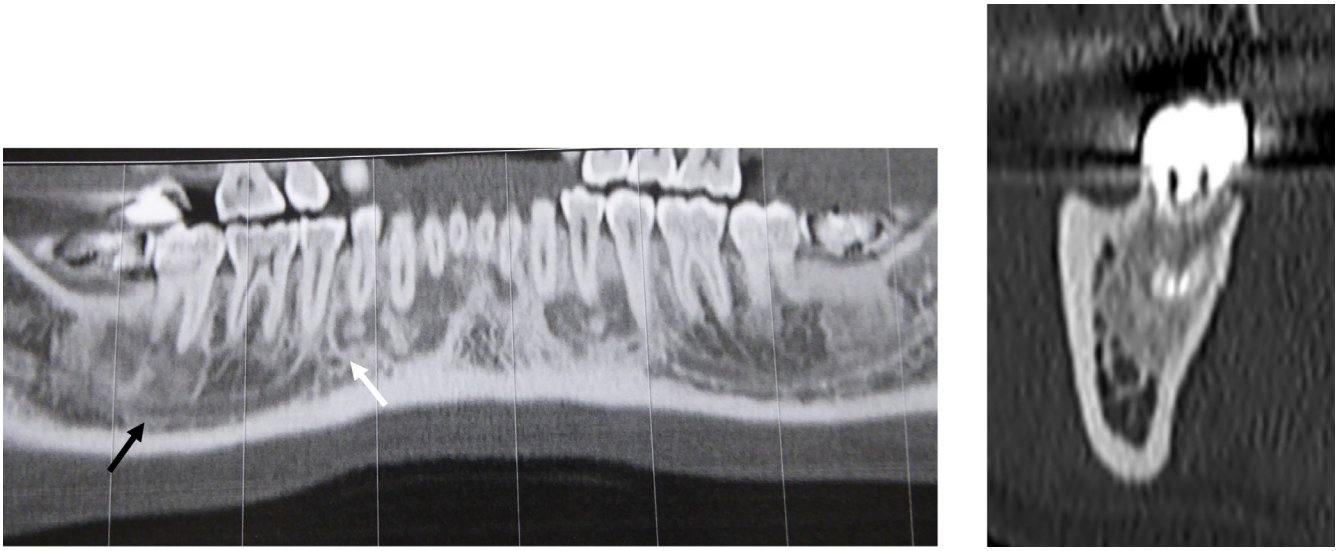

Fig. 3. CBCT initial scan: irregular radiopacity lesions surround the apical region of tooth 47 (black arrow) and near by the inferior alveolar nerve foramen (white arrow). (a) Coronal cut. (b) Sagittal cut near by the tooth 47.

close contact with the mental foramen and the right mandibular foramen (Fig. 3a and b). These lesions were bilateral and symmetrical, multilobulated, irregular in shape, of mixed appearance, and had clear margins. Florid osseous dysplasia was diagnosed on the basis of these scans. Conservative monitoring was established on an annual basis. Six month later, the patient consulted after the occurence of paresthesia as burning and tingling sensation and pain in the right labiomental region. Yet, any infection of the lesions were observed. Following the altered symptomatology, a root canal treatment of the teeth [5] in contact with the lesions was performed under antibiotic prophylaxis then a resection of the sequestered cementoosseous was done under general anesthesia (Fig. 4). The panoramic X-ray performed following the surgery confirmed the complete resection of the lesions (Fig. 5). The pathological examination confirmed the diagnosis of cementomas with the presence of irregular pagetoid osteoids of different shapes, sizes, and thicknesses with irregular cement lines. Osteoblasts were present in contact with the newly formed bone. Bone segments were filled with benign fibroblast proliferations. One month after surgery, the patient showed good wound healing, the neuropathic pain had disappeared but there was still an isolated numbness of the oral mucosa compared to the controlateral area. Six months after surgery, the hypoesthesia had almost disappeared; the panoramic X-ray showed a gradual filling of cavities and a lack of recurrence (Fig. 6).

Comments

Florid osseous dysplasia is a rare and benign condition characterized by a multifocal onset that can affect all four quadrants of the mouth. It mainly affects women of African origin between the ages of 40 and 50 years [6,7]. Although the etiology and the triggering factors were not identified [8], Florid osseous dysplasia could result from abnormal activity of the periodontal ligament explaining its near-exclusive location in the dental region, with cemento-osseous lesions identified in relation to dental apices and dental anatomy in radiographic examinations [9]. It is generally asymptomatic and the teeth in relation to the lesions are unaffected

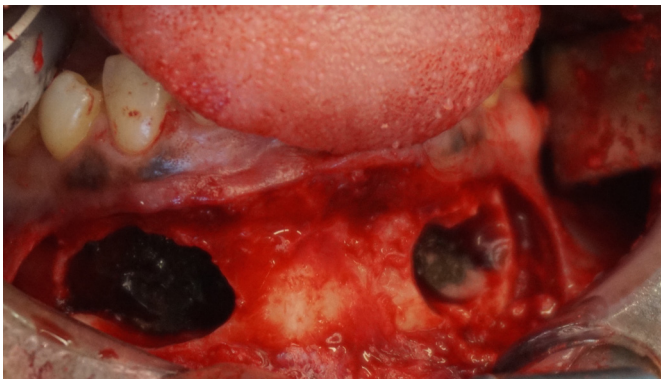

Fig. 4. Bone cavities after osseous dysplasia resection.

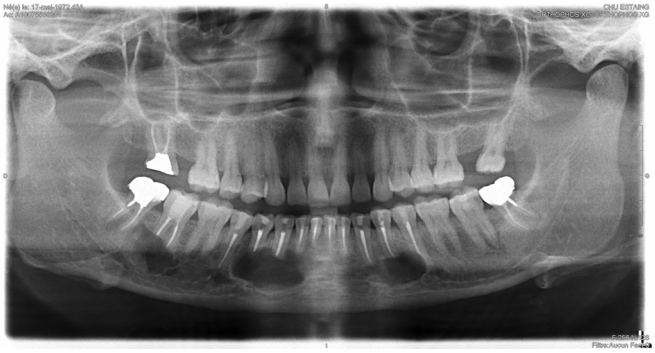

Fig. 5. Post-operative panoramic radiography.

[10–12]. Florid osseous dysplasia becomes symptomatic when lesions are accompanied by infection, or if cortical fenestrations occur [3]. The diagnosis is radiological with the presence of mixed multilobular well-defined images of the alveolar bone, respecting the basilar bone. Any rhizalysis are reported [11]. Other diagnoses are ossifying fibroma, Paget disease, diffuse chronic osteomyelitis, Gardner syndrome, and endodontic lesions [12]. In the absence of symptomatology, it is recommended to conduct annual clinical and radiological monitoring and to maintain rigorous oral hygiene to avoid infections [13]. However, because of their sclerotic and vascular nature, lesions are particularly susceptible to infection after mucosal trauma, dental procedures, biopsy or cystic enucleation [14,15]. This risk of iatrogenic infection can justify an antibiotic prophylaxis in any surgical procedure [16]. The infection is accompanied by soft-tissue swelling and pain but without any hypoesthesia in the territory of the inferior alveolar nerve [13,15,17–19].

In the presented case, the pain has occured without any infectious process or mucosal lesion. The symptoms are typical of neuropathic pain with burning sensation, tingling and electric shocks of the lower lip. According to the Kawai classification, CBCT unveiled stage-E lesions (irregular radio-opaque lesions surrounding the dental root) and stage-D lesions (lobular or spherical calcified masses surrounded by a radiolucent border) around the roots of the teeth 46, 47, and 48, which is typical of florid osseous

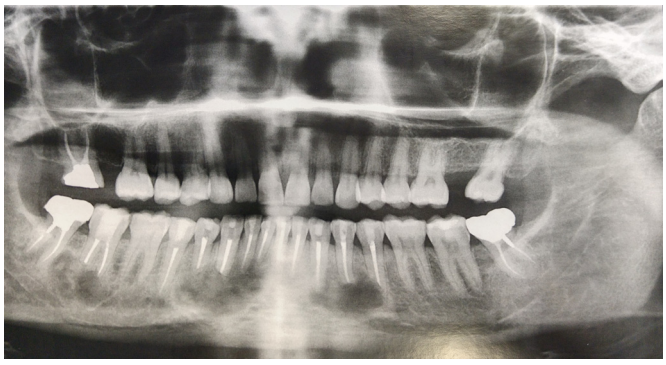

Fig. 6. Six months post-op panoramic radiography: a gradual mineralization of the intra-osseous cavity is obvious.

dysplasia [20]. The proximity between the calcified masses and the mental canal or mandibular foramen could justify the sensitive disorders.

In the absence of bony sequestrum, surgery is difficult because the lesions can not be easily separated from the healthy bone and its is necessary to perform curettage of the alveolar bone reach healthy bone [21]. Some authors recommend that cortical fenestrations be performed in lesions to induce better healing of bone tissue [22]. When faced with the cleavable aspect of the apical cemento-osseous lesions and the need for a diagnosis, we opted for a surgical resection of the lesions and a grinding of the cavity walls after enucleation.

Despite a lack of consensus regarding this, in cases of multifocal lesions in the presence of decayed or treated teeth or the risk of pulp necrosis or infection after a surgery, endodontic treatment of all the teeth in contact with the lesions is recommended. The two procedures (endodontic treatment and lesion resection) involved an antibiotic prophylaxis because the literature reported a case of osteomyelitis after an endodontic treatment in a patient with florid osseous dysplasia [23].

Conclusion

There is no consensus on the management of florid osseous dysplasia. Nevertheless, some guidelines are found in the literature. If the patient is asymptomatic, clinical and radiological follow-up must be instituted. If the patient has an infection, an antibiotic treatment and sometimes a resection of sequestered cemento-osseous apices should be proposed. In case of neuropathic pain, the anatomical decompression of the lower alveolar nerve relieves the pain.

Conflicts of interests: The authors declare that they have no conflicts of interest in relation to this article.

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