

Article

SURGICAL MANAGEMENT OF OSSIFYING FIBROMA IN THE MAXILLA AND ITS CHALLENGES ASSOCIATED WITH AN IMMEDIATE RECONSTRUCTIVE APPROACH WITH AUTOGENOUS GRAFT, AN UNUSUAL CASE REPORT

Manejo quirúrgico del fibroma osificante en el maxilar y sus desafíos asociados con un enfoque reconstructivo inmediato con el injerto autógeno, un informe de caso inusual

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Receipt: 2021/03/27
Acceptance: 2021/06/04

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ABSTRACT

Ossifying Fibroma is a gingival mass in which calcified foci are found. It is a nodular lesion that involves the inserted and interdental gum. There is a predilection for the female sex and for the anterior region of the maxilla. This work aims to report the surgical management of a Ossifying Fibroma in the maxilla and the immediate reconstructive with autogenous graft. Female patient, in the third decade of life, presenting Ossifying Fibroma in the region between maxillary incisors and premolars. She was treated surgically by means of marginal resection of the lesion under general anesthesia and immediate reconstruction with autologous iliac crest graft. Followed up for 12 months with no signs of recurrence.

Keywords: Ossifying fibroma; Jaw; Bone graft; Iliac crest.

1. Introduction

Ossifying Fibroma is part of fibro-osseous lesions, which is a group of benign pathologies involving the jaws (Mafee, *et al.*, 2003). The first description of this pathology was made by Shepherd and collaborators in 1844 (Franco-Barrera, *et al.*, 2016). The term Ossifying Fibroma was given by Eversole and Rovin in 1972 (Rallan, *et al.*, 2013).

Ossifying Fibroma has an unknown etiology, being more frequent between the third and fourth decade of life and with a predilection for females (Mafee *et al.*, 2003). Among the etiological factors associated with this pathology are mutations related to multipotent cells of the periodontal ligament, also mutations in the gene HPRT2 and there is an assumption that it would be some stimulus induced by previous extraction and periodontitis or simply be linked to a disturbance in bone maturation of congenital origin (Khan *et al.*, 2011; Liu *et al.*, 2010; Mohsenifar *et al.*, 2011).

Clinically, it presents as a small focal mass well demarcated in the gingiva with a sessile or pedunculated base, usually originating from an interdental papilla (Rallan, *et al.*, 2013). It presents a slow and locally aggressive growth, causing facial asymmetry in cases of long evolution (Mohsenifar *et al.*, 2011). The lesion is asymptomatic until it affects some vital structure in its vicinity, such as the mandibular canal, nasomaxillary region and orbit,

causing paresthesia of the lower alveolar nerve in the lower third of the face and epistaxis, epiphora, dystopia and diplopia when it reaches the middle third of the face (Mafee *et al.*, 2003).

The lesion affects both the maxilla and the mandible. The occurrence in the maxilla is up to 60%, 50% of which occur in the anterior region of the maxilla (Franco-Barreira *et al.*, 2016). In the posterior mandibular region is affected in 70 to 90% of cases (Liu *et al.*, 2010). The diagnosis of the lesion usually occurs on routine radiographic exams, as it presents a slow and asymptomatic evolution (Mainville *et al.*, 2017). The differential diagnosis of Ossifying Fibroma includes fibrous hyperplasia, pyogenic granuloma giant peripheral cell granuloma and peripheral odontogenic fibroma (Gomes *et al.*, 2019). The final diagnosis of this type of lesion is usually made by histopathological examination (John *et al.*, 2016). The treatment of choice is total enucleation and the removal of possible causative agents (Parihar *et al.*, 2015).

The aim of this article is to report a peculiar case of a young patient, who was diagnosed with Ossifying Fibroma in the maxilla, where he was surgically approached with the complete removal and immediate reconstruction of the bone defect generated with autologous iliac crest graft.

2. Case report

Female patient, 25 years old, melanoderma was referred to the specific care program for stomatological diseases, Federal University of Uberlândia, Uberlândia, Brazil. After a routine visit to the dentist, due to a radiographic finding in the right region of the maxilla. In the anamnesis, the patient did not report any comorbidities, allergies or medical treatment in progress.

On clinical extra-oral examination, the patient did not present asymmetry on the face. The intraoral examination showed a bulging in the bottom of the right vestibule between the dental elements 12 to 15, with a hardened consistency, without pain on palpation and absence of tooth mobility.

Panoramic radiographic examination (Figure 1) shows a radiopaque lesion in the region of elements 12 to 15, with severe tooth resorption of elements 13 and 14 and root divergence of elements 12 and 15. In cone beam computed tomography (Figure 2), the extent of the lesion was observed with proportions of 18.13 mm mesio-distal length; 15.19 mm in vestibule-palate length; 19.12 mm in corono-apical length and with complete resorption of the roots of elements 13 and 14.

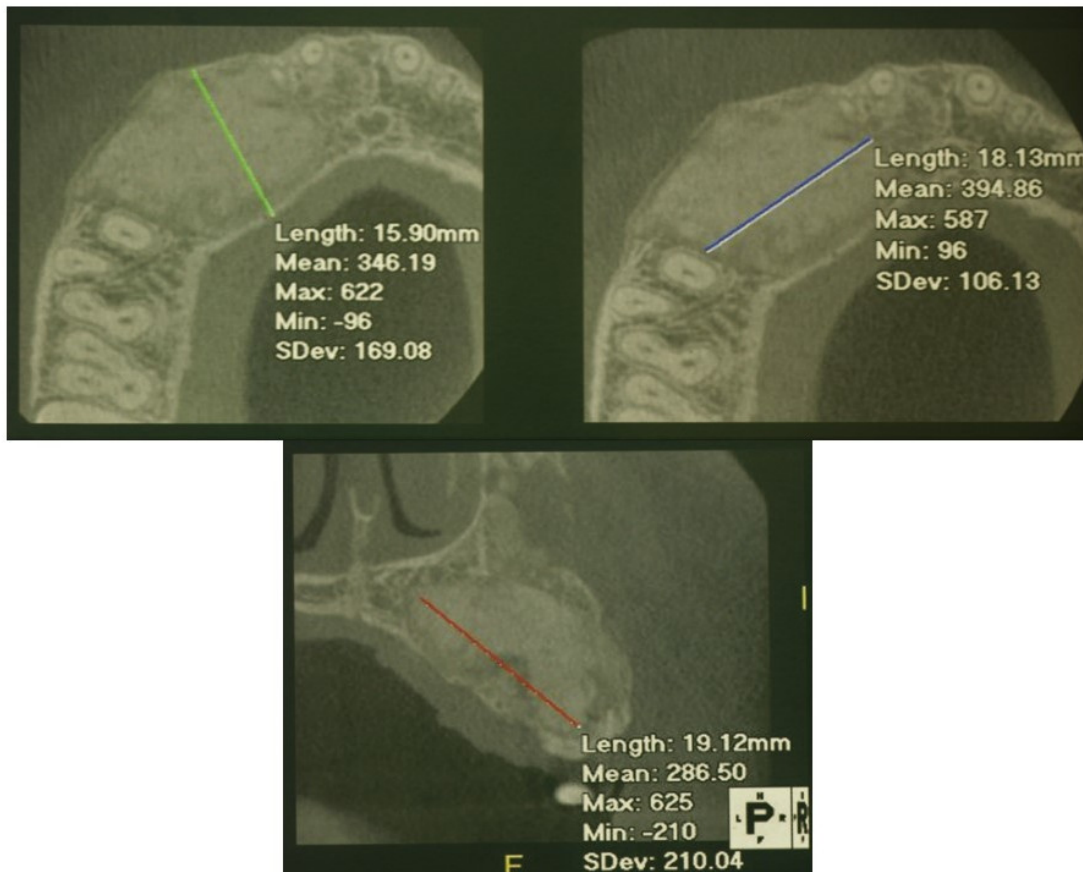
Figure 1.

Initial panoramic radiograph, showing lesion in the right maxilla.



Figure 2.

Cone beam computed tomography, showing the lesion proportions in the right.



The diagnostic hypotheses given were osteoma, ossifying fibroma and myxoma.

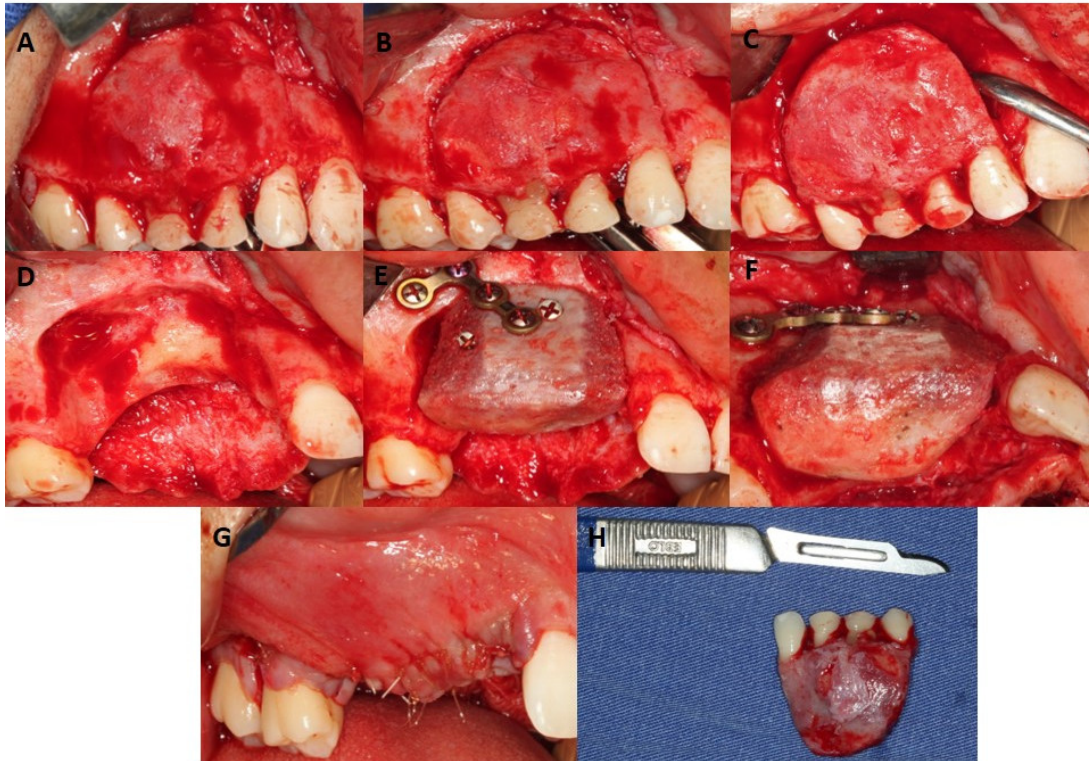
Under local anesthesia, an incisional biopsy was performed through an intraoral approach to confirm the diagnosis. The material collected from the surgical bed was sent for histopathological analysis, which concluded to be an ossifying fibroma. After confirmation of the diagnostic hypothesis, the patient underwent marginal resection of the lesion under general anesthesia and immediate reconstruction of the defect generated, with autogenous crest graft iliac.

Anesthetic induction was performed, with tracheal intubation. Extraoral antisepsis was performed with 2% chlorhexidine and intra-oral with 0.12% chlorhexidine. The surgical bed was infiltrated with 0.5% bupivacaine with epinephrine 1: 100,000, for hemostasis during the procedure. An intraoral mucous-periosteal flap was performed, exposing the entire region affected by the lesion (Figure 3A). With a no. 702 drill, the resection margin was delimited (Figure 3B) and later with chisels the complete removal of the bone block (Figure 3C). Then with the maxicut drill, the osteotomy was performed for regularization and removal of unfavorable contours (Figure 3D).

Afterwards, the orthopedics team performed the removal of the tri-cortical iliac crest graft, for immediate reconstruction of the maxillary defect generated by the resection. The graft was then prepared for better adaptation in the recipient bed and thus fixed with a titanium miniplate of the 2.0 system, and 2 10mm long screws, allowing adequate graft stabilization (Figure 3E-3F). Consequently, the operated region was abundantly irrigated with saline and the flap was closed completely with resorbable threads, without any exposure of the graft inside the mouth (Figure 3G). Resected anatomical specimen (Figure 3H).

Figure 3- 3A

Muco-periosteal flap exposing the entire lesion in the maxilla region; 3B Marking the lesion resection margin; 3C Mobilization of the block after marginal resection; 3D Surgical bed after removal of the resected marginal block and regularization osteotomy performed; 3E Positioning of the iliac crest graft in the surgical bed, in the frontal view; 3F Positioning of the iliac crest graft in the surgical bed, in the alveolar view; 3G Muco-periosteal flap closure, without graft exposure in the intraoral environment; 3H Resected anatomical specimen.

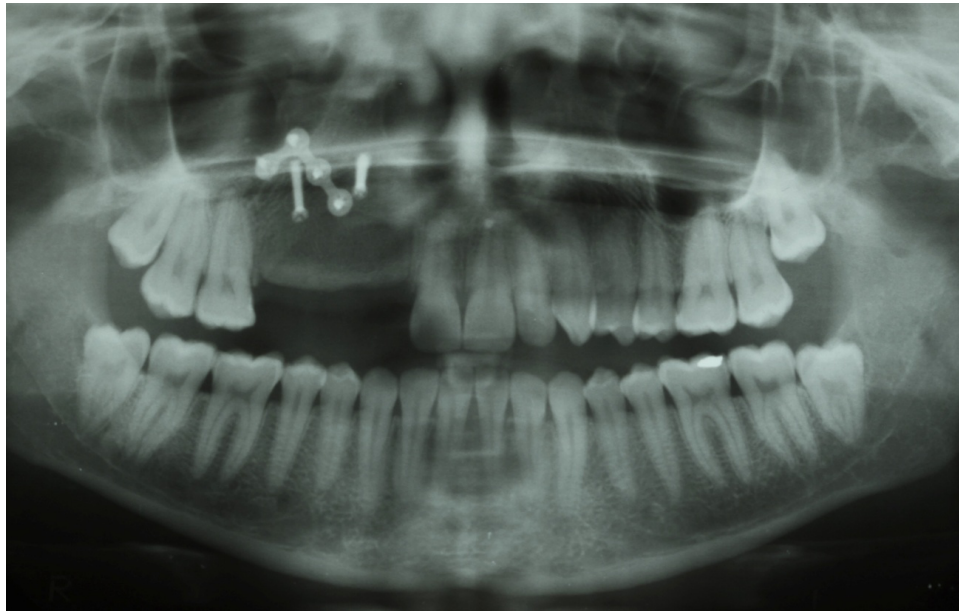


Due to the size of the graft used in maxillary reconstruction, the patient underwent hyperbaric oxygen therapy sessions, in a hyperbaric chamber with 1.5 isobaric phases, for a total of 30 postoperative sessions (lasting 90 minutes each session at speed 006 to 012 KJF /cm²/ minute), for better predictability of surgical success.

After hospital discharge, the patient was medicated with amoxicillin 500mg 3x a day for seven days, dexamethasone 4mg 2x a day for four days, toragesic 10mg 3x a day for 4 days and a mouthwash of 0.12% chlorhexidine digluconate 0.12% 2x a day for ten days. On weekly outpatient visits to the patient, she had no complaints, no dehiscence of the surgical wound and no phlogistic signs. The patient was followed up on an outpatient basis for 12 months with quarterly radiographic follow-ups (Figure 4), being discharged after this period, for dental rehabilitation.

Figure- 4

Final panoramic radiograph after 12 months of surgical procedure.



3. Discussion

Ossifying Fibroma is a lesion that frequently affects women (Kale *et al.*, 2014), in the present case a woman in her third decade of life was affected. Studies show that the pathogenesis of the lesion is associated with local irritating factors, such as dental plaque, calculus, microorganisms, masticatory forces, poorly adjusted dental prostheses and low-quality restorations have been implicated in the etiology of Ossifying Fibroma (Pal *et al.*, 2012). Hormonal factors are associated with a higher occurrence in women (Kenney *et al.*, 1989).

Clinically, Ossifying Fibroma appears as an exophytic mass adhered to the gingiva, and this ulcer may share characteristics like other extraosseous lesions, such as pyogenic granuloma, peripheral giant cell granuloma (Das UM & Azher, 2009). Despite ossifying fibroma showing benign behavior, asymptomatic and slow growth, when left untreated, its evolution can cause critical damage to patients (John *et al.*, 2016).

Radiographically, depending on the size of the ossification foci, radiopaque spots can be seen on routine radiographs, such as the periapical and panoramic ones (Marcos, *et al.*, 2010), which are usually unilocular and well defined.

Histopathologically, the lesion may present with frequent epithelial ulceration, with the presence of plump or spindle cells, with small nuclei and poorly defined cytoplasm. It may present mineralization showing different combinations of immature, mature bone (lamellar), cementum or calcifications (Lázare *et al.*, 2009).

The treatment of choice is local resection with peripheral margins, removal of the periodontal ligament and affected periosteal component. In addition, it is necessary to eliminate local etiological factors, such as bacterial plaque and calculus (Kale *et al.*, 2014). Surgical excision of the lesion becomes the treatment of choice in these cases due to the significant reduction of recurrences, when compared to conservative treatment through enucleation and removal of the periosteal tissue adjacent to the lesion (Emerson *et al.*, 2012). As in the case described, marginal resection of the injury with safety margins was the treatment of choice for this case, with an extremely satisfactory evolution (Carvalho *et al.*, 2012; Kharsan *et al.*, 2018). However, radical treatment can cause severe sequelae to patients, requiring more procedures so that functional and aesthetic rehabilitation is achieved, the that was not necessary in the reported case (Emerson *et al.*, 2012).

In the present case, after resection there was a need to reconstruct the bone defect immediately. The great defects caused by resections of neoplasms the iliac crest is the most common donor site for the collection of autogenous grafts. The anterior iliac crest provides bone volume between 30 and 50 ml and the posterior iliac crest between 60 and 80 ml (Mazzonetto, 2009). Among the advantages of the autogenous graft, we can mention its immunocompatibility properties and the osteoinductive, osteogenitizing and osteoconductive capabilities, guaranteeing a rich cellularity and potential for early revascularization (Marx, 2007). Thus, reconstruction for later rehabilitation is more predictable.

Among the disadvantages of iliac crest grafts, we can mention its higher rate of resorption, due to the greater part of its microstructure being spongy, and some comorbidities related to the graft donor site, such as chronic pain, sensory loss, hematoma and hemorrhage, joint instability sacroiliac and gait disorders (Nkenke *et al.*, 2002; Kao & Scott, 2007). In order to optimize the bone neoformation process of the autogenous graft, hyperbaric oxygen therapy becomes a good option.

Several studies have already shown its efficiency when used in conjunction with the grafting procedure. Among its effects that favor bone neoformation, we can mention a better vascular supply, favoring osteoblastic activity during the bone remodeling process, increased oxygen tension in tissues, facilitating the proliferation of fibroblasts and osteogenesis and the facilitation of polymorphonuclear leukocyte function (Hunt *et al.*, 1969; Hunt & Pai, 1972). All these mechanisms, when favored by the increase in oxygen pressure in the tissues, culminate in a better process of tissue repair and regeneration, decreasing the degree of absorption of the bone graft and accelerating its incorporation to the receptor site (Gill & Bell, 2004).

As mentioned in the report, the adoption of hyperbaric oxygen therapy proved to be of unparalleled importance in relation to the final success of the treatment, making the rehabilitation receptor bed (iliac crest graft block) more appropriate, since bone integration and osteoinduction levels were achieved successfully.

Ossifying fibroma, despite being widely referenced in the world literature, is still a pathology that poses challenges to surgeons, especially about high rates of recurrence when conservative treatments are adopted. In the case described, it proved to be an enormous challenge, since the affected region was unusual, the maxilla, with the adoption of a resection treatment with a safety margin, associated with bone reconstruction with autologous iliac crest graft together with adjuvant hyperbaric oxygen therapy, for increased case predictability. In the end, it can be said that the result obtained is within satisfactory, and in this case, it shows an unusual approach regarding the treatment plan adopted.

4. Ethical aspects:

There is compliance with ethical standards through informed consent on the publication of the article.

5. Conflicts of interest:

There is no conflict of interest between the authors.

6. Fundings:

There is no funding.

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RESUMEN

El Fibroma osificante es una masa gingival en la que se encuentran focos calcificados. Es una lesión nodular que involucra la encía adherida e interdientaria. Hay una predilección por el sexo femenino y por la región anterior del maxilar. Este trabajo tiene como objetivo informar sobre el manejo quirúrgico de un fibroma osificante periférico en el maxilar y su reconstrucción inmediata con injerto autógeno. Paciente de sexo femenino, en la tercera década de vida, presenta un Fibroma osificante en la región entre incisivos maxilares y premolares. Fue tratada quirúrgicamente por medio de una resección marginal de la lesión bajo anestesia general y reconstrucción inmediata con injerto de cresta iliaca autóloga. Seguimiento durante 12 meses sin signos de recurrencia.

Palabras clave: fibroma osificante; hueso maxilar; cresta iliaca; injerto óseo.
