Revista 16 de abril. 2023; 62:e1625
Case presentation

Ossifying fibroma of the upper maxilla: A case report

Fibroma osificante de maxilar superior: informe de un caso

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ABSTRACT

Ossifying fibroma is a benign osteogenic mesenchymal tumor. It is presented the case of an 18-year-old female patient, with black skin, who attended the maxillofacial surgery clinic due to an increase in nasal and upper jaw volume on the left side, slow-growing, asymptomatic. The panoramic X-ray confirmed a well-defined unilocular radiolucent image, with displacement of tooth roots without root resorption. The biopsy confirmed a neoplastic connective tissue stroma, with the presence of bone spicules with a laminar peripheral structure. The diagnosis was ossifying fibroma of the upper jaw. Complete resection of the tumor was performed, including teeth related to it, and the bone defect was repaired with hydroxyapatite. The postoperative evolution was favorable. This condition requires multidisciplinary management. Its diagnosis is a clinical one, supported by complementary tools such as imaging and histopathology. It is a very rare benign tumor that occurs in young people. It is important to make a diagnosis in order to differentiate it from other fibrous lesions.

Keywords: Ossifying fibroma; Maxillary; Neoplasm.

RESUMEN

El fibroma osificante es un tumor mesenquimatoso osteogénico benigno. Se presenta el caso de una paciente de 18 años, de piel negra, que acude a consulta de cirugía maxilofacial por presentar aumento de volumen nasal y maxilar superior del lado izquierdo, de crecimiento lento, asintomático. La radiografía panorámica constató imagen radiolúcida unilocular bien definida, con desplazamiento de raíces de dientes sin reabsorción radicular. La biopsia comprobó un estroma de tejido conjuntivo de constitución neoplásica, con presencia de espículas óseas de estructura periférica laminar. El diagnóstico fue de fibroma osificante de maxilar superior. Se realizó resección completa del tumor, incluyendo dientes relacionados con el mismo y reparando el defecto óseo con hidroxiapatita. La evolución posoperatoria fue favorable. Esta afección requiere un manejo multidisciplinario, diagnosticada con la clínica, apoyada en herramientas complementarias como la imagenología e histopatología. Es un tumor benigno muy raro que se presenta en personas jóvenes. Es importante realizar un diagnóstico con el fin de diferenciarlo de otras lesiones fibrosas.

Palabras clave: Fibroma osificante; Maxilar; Neoplasia.

OPEN ACCESS **Published:** January 5th, 2023 || **Received:** March 8th, 2022 || **Accepted:** May 13rd, 2022

Cite as:
Leyva-Brooks S, Ballart-Ramos A, Román-Luna D. Fibroma osificante de maxilar superior: informe de un caso. Revista 16 de abril [Internet]. 2022 [cited: access date]; 62(287):e1625. Available from: http://www.rev16deabril.sld.cu/index.php/16_04/article/view/1625

INTRODUCTION

Benign fibro-osseous lesions of the craniofacial mass represent a group of alterations in which there is replacement of normal bone by fibrous connective tissue. Although they present different clinical characteristics, their radiographic and histologic features are similar. Their prognosis and treatment depend on this[1].

These lesions were classified by Waldron and Giansanti into three groups: fibrous dysplasia, fibrous lesions that appear in the periodontal ligament (central ossifying fibroma and peripheral ossifying fibromas) and fibro-osseous neoplasms of uncertain or debatable relationship (osteoblastoma and cementumblastoma)[2]. Benign tumors include sinonasal papillomas, juvenile nasopharyngeal angiofibroma and fibro-osseous lesions[3]. Ossifying fibroma, the rarest of these entities, represents a true neoplasm[4, 5].

In 1872 Menzel was the first to describe a fibro-osseous lesion; however, it was not until 1927 that the term "ossifying fibroma" was used for the first time by Montgomery, the name by which the lesion is currently recognized. Until 1948 it was thought that fibrous dysplasia and ossifying fibroma were the same disease or that one was a variant of the other. It was in the early fifties of the 20th century that Sherman and Sternberg, with their detailed studies of the clinical, radiological, histological and pathological aspects, divided them into two different identities. The name "ossifying fibroma" was adopted in the early 1990s by the World Health Organization (WHO)[6].

It affects the jaws and long bones. It is characterized by being encapsulated and well delimited and presenting expansion of the cortical lamina, in addition to deformity of the affected bony structures. It also affects craniofacial structures. It should be noted that some authors consider it to be a proliferative lesion, which expands slowly and progressively. In addition, clinically it is a benign neoplasm that affects with greater continuity the mandibular bone posterior to the canine region and, occasionally, the maxilla or other bones of the skull. It is especially prevalent in females due to the vascular effects of hormones[7].

Ossifying fibroma (OF) is a benign osteogenic mesenchymal osteogenic tumor originating from the periodontal ligament. It is intraosseous, asymptomatic, with aggressive behavior. It is usually large in size, significantly destroys the affected bone and tends to recur in 30 % to 58 % of cases. It is capable of producing a great variety of benign neoplasms that jeopardize the integrity of the bone cortices, generating expansion and progressive destruction. Some authors relate the appearance of FO to irritant factors, bacterial plaque and subgingival calculus[1,8]. Triggering factors for the development of this neoplasm are associated with exodontic trauma and periodontal disease[8]. It has been determined that FO has a prevalence in Latin America of 0.1% of all benign cementum-osseous conditions[9].

Due to the low frequency of this type of lesion, knowledge of its diagnosis and treatment is relevant, so this article describes the clinical-surgical management of a patient with OF in the upper jaw.

CASE PRESENTATION

Female patient, 18 years old, black skin, urban origin, with personal pathologic history of bronchial asthma and regular treatment with prednisone, who came to maxillofacial surgery for presenting nasal and maxillary enlargement on the left side, with slow growth, asymptomatic and with one year of evolution, with no history of trauma in the area.

The extraoral physical examination revealed facial asymmetry, an increase in volume of 5 cm in diameter, of hard stony consistency, extending to the infraorbital region and the maxillary area on the left side, without sensory compromise, with effacement of the nasolabial fold and elevation of the nasal wing (fig. 1).

The intraoral physical examination revealed a firm, well-defined mass with involvement of the left maxillary maxillary hemiarch from the canine to the second premolar with effacement of the vestibular sulcus, bulging of the vestibular and palatal cortex without fistulas or intra- or extraoral drainage, without cervical adenopathies, and with a slight change in the occlusal plane in the posterior sector (fig. 2).

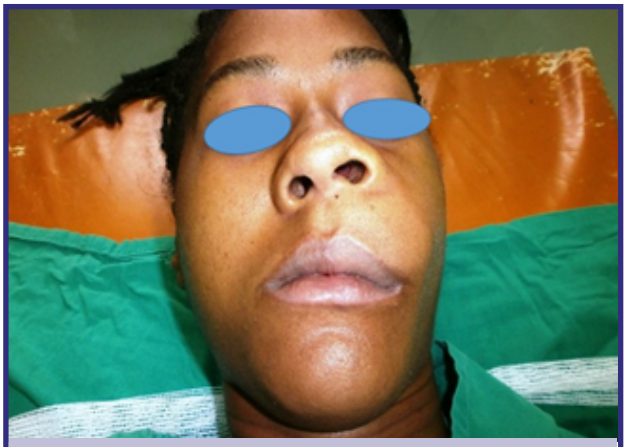

Fig. 1. Anteroposterior view of the patient at facial physical examination

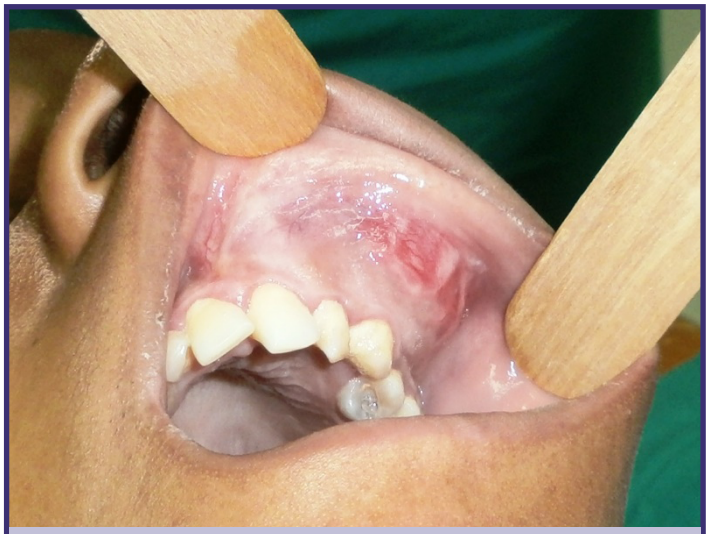

Fig. 2. Intraoral physical examination

Analysis of the panoramic radiograph confirmed a well-defined unilocular radiolucent image, with displacement of the roots of the teeth without root resorption. On Walter view a radiolucent area was observed in the left maxilla involving the nasal region and invading the maxillary sinus.

An incisional biopsy was performed under local anesthesia, in which a tissue of hard consistency was obtained, which was subsequently subjected to histopathological study, where a cream-colored, yellowish homogeneous mass was observed on section. The lesion was formed by a connective tissue stroma of neoplastic constitution, with the presence of bone spicules of lamellar peripheral structure, surrounded by rows of osteoblasts. No anaplasia or necrosis was found; the edges of the lesion presented a thin layer of compressed bone. The diagnosis of maxillary OF was thus confirmed.

Complete resection of the tumor was performed under general anesthesia (fig. 3), including all related teeth and repairing the bone defect with hydroxyapatite. However, the first and second molars, which were displaced but not involved in the lesion, were not removed. The vestibular and palatal mucosa was preserved to obtain closure by first intention.

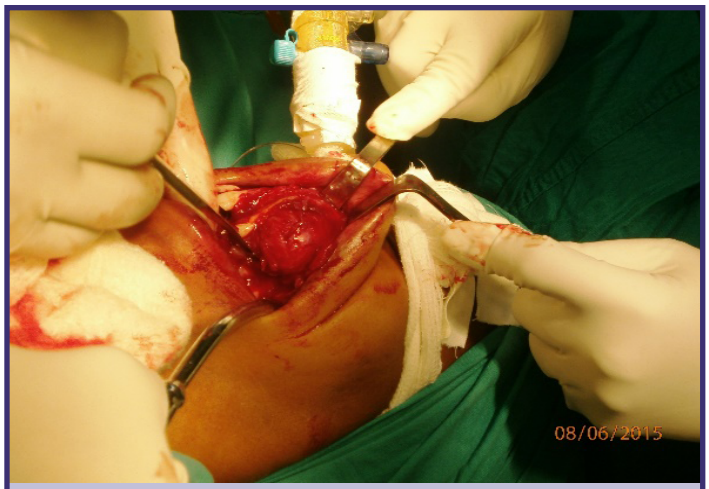

Fig. 3. Postoperative course

Postoperative evolution was favorable both clinically and radiographically - the latter was noted weekly for one month (fig. 4), then monthly for three months and then every three months for one year. Progressively, the patient was referred to the prosthodontics specialty for rehabilitation of the extracted teeth.

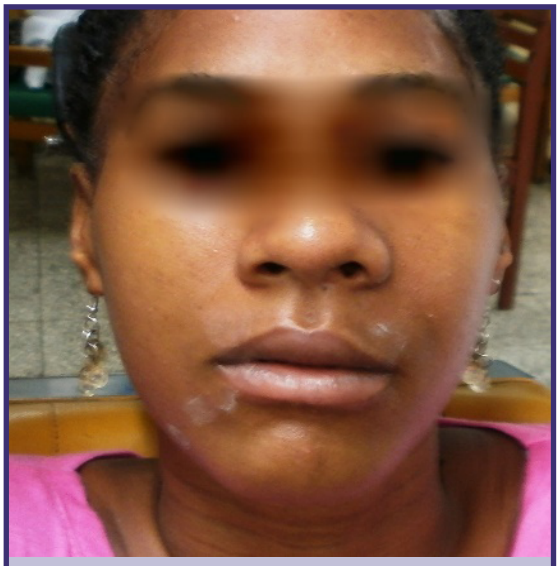

Fig. 4. Evolution 15 days after surgery

DISCUSSION

The WHO classification of head and neck tumors (2017) divides OF into fibro-osseous and osteochondromatous lesions. According to age of onset, clinical presentation, potential behavior, mineralization pattern and histopathological features, three variants have been identified: cementum-OF (COF) and two distinct juvenile OFs (JOF) [juvenile psammomatoid OF (JPOF) and juvenile trabecular OF (JTOF)][10], hence the clinical case is classified according to the latest WHO update.

Lopez JA *et al.*[7] determined important clinical features that point to the benign behavior of the tumor, such as the slow and gradual growth of the lesion, as well as the area of volume increase that presented covered by mucosa of normal characteristics to the adjacent tissue, results similar to this case.

Kawaguchi *et al.*[10] state that cementum OF has been reported most frequently in adults between 16 and 33 years of age, which coincides with the patient's age.

It affects the mandible in 70% to 80% of cases and then preferentially localizes to the premolar-molar area, followed by the maxilla and paranasal sinuses. OF is an uncommon lesion and few case series have been published, mainly in the maxillary region[2], hence the peculiarity of this case.

Several related articles[1,7,8] state that histopathologic examination and radiographic examination are required to obtain a definitive diagnosis of the entity, which was adapted in this case.

Viana *et al.*[11] describe that enucleation or curettage of the lesion is the treatment of choice for cementum-ossifying fibroma, as it was in this case.

The differential diagnosis depends on the degree and pattern of internal radiopacity. It includes radiolucent and radiopaque mixed benign neoplasms, with the diagnosis determined by clinical and radiographic behavior[12]. Fibrous pathological entities resembling OF are: osseocemental dysplasia (focal periapical and florid dysplasia), fibrous dysplasia, osteoid osteoma, osteoblastoma and chronic sclerosing osteomyelitis[13]. Other possible options for differential diagnosis are: solitary bone cyst, keratocyst, ameloblastoma, giant cell granuloma, myxoma, adenomatoid odontogenic tumor, multiple myeloma, osteosarcoma and cementoblastoma[14,15].

At present, COF continues to be a subject of study and discussion. This condition requires multidisciplinary management and should be diagnosed primarily on clinical grounds, supported by complementary tools such as imaging and histopathology to provide a definitive diagnosis.

CONCLUSIONS

OF is a very rare benign tumor that occurs in very young people. It is important to make a proper diagnosis in order to differentiate it from other fibrous lesions to allow early appropriate therapy.

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CONFLICT OF INTERESTS
Authors declare there was no conflict of interests.

AUTHORSHIP
Santiago Leyva-Brooks: conceptualization, content and data curation, investigation, metodology, writing-original draft.
Aliandri Ballart-Ramos: conetn and data curation, metodology, writing-original draft.
Daimeris Román-Luna: conceptualization, investigation, metodology, supervision, writing-original draft.

FUNDING
No funding was received for this research's development.