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Case Report

Ossifying Fibroma of the Maxilla in Two Cases and Review of the Literature

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Abstract: Ossifying fibroma is a benign tumor that rarely affects the face bones and exclusively the maxilla. We report two cases of ossifying fibroma of the maxilla treated in our department of maxillofacial surgery and stomatology at IBN SINA university hospital in Rabat. The 1st case is a 38-year-old female, with no particular history, who presented a progressive swelling of the left face since 05 months. Extraoral examination demonstrated a grade I left exophthalmos, without diplopia or limitation of eye movements. Intraoral examination revealed a vestibular expression of the tumor. The 2nd case is also a 45-year-old female, who presented a progressive swelling of the left maxilla which gradually increased in size over a period of 04 months. Intra-orally, the buccal vestibule was completely obliterated with the mass. The two patients presented radiological images of similar density, made of a heterogeneous tissue of the maxillary containing calcifications but with different extension. Surgical excision was performed in both patients and the diagnosis was confirmed by histological study of the resected specimen. The evolution of the ossifying fibroma is insidious; this pathology has a clinical polymorphism and a good prognosis. The etiopathogenesis of this tumor is still unknown, it's more likely to develop in the mandible and very rare in the maxilla. It differs from other types of fibroma by their clinical, radiological and histological aspects. However, only the pathological examination confirms the diagnosis.

Keywords: Ossifying fibroma, the maxilla, orbital floor reconstruction.

INTRODUCTION

Ossifying fibroma is a benign tumor that rarely affects the bones of the face and exclusively the maxilla. Ossifying fibroma was first described by MENZEL in 1872 and then in 1927 by Montgomery [1] and since rare are the cases of ossifying fibroma reported in literature originating in the maxilla.

The evolution of ossifying fibroma is slow and generally asymptomatic, unless it causes facial deformity or functional discomfort. It is a tumor with a good prognosis, but it can have an aggressive character [2, 3].

CASE REPORT

In this article, we report two cases of ossifying fibroma of the maxilla treated in our department of maxillofacial surgery and stomatology at IBN SINA university hospital in Rabat.

Case N ° 1: A 38-year-old female, with no particular history, the patient presented a progressive swelling of the left cheek for 05 months. Extraoral examination demonstrated a grade I left exophthalmos, without diplopia or limitation of eye movements [fig. 1. A]. Intraoral examination revealed a vestibular expression of the tumor.

The patient underwent a CT scan of the face, which revealed a heterogeneous tissue process in the left maxillary sinus that enhanced after injection of the contrast product, containing calcifications and measuring 2.6cm x 4 cm. This process destroyed the orbital floor, blows the side wall of the ethmoid and respects the pterygoid region and the base of the skull [fig.2].

The patient was scheduled for surgery; we did a vestibular incision and resected the whole tumor from

there. The orbital floor was invaded and resected; the reconstruction was made by a graft of the iliac crest through a transconjunctival approach [fig.3].

Anatomopathological study confirmed the diagnosis of the maxillary ossifying fibroma. The postoperative follow-up was simple. There was no facial asymmetry, no enophthalmos, no diplopia and no limitation of the eye movements, on a follow-up of 03 years [fig.1.B] [fig.4].

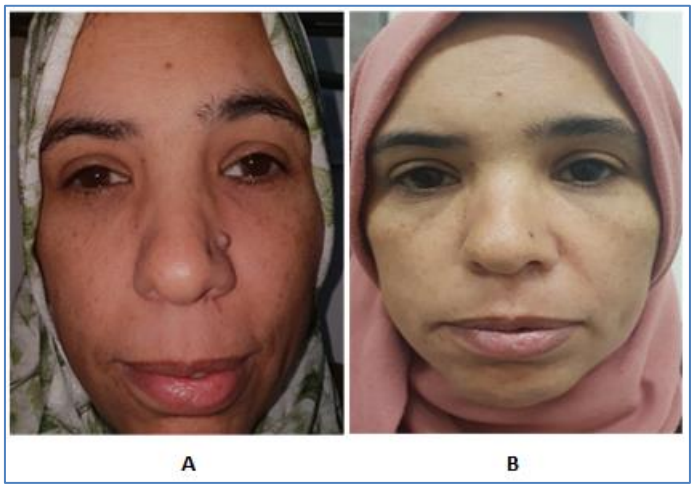

Fig-1: A: preoperative picture showing a left cheek swelling with exophthalmos of the homolateral eye classified grade I. B: picture taken 2 years after surgery

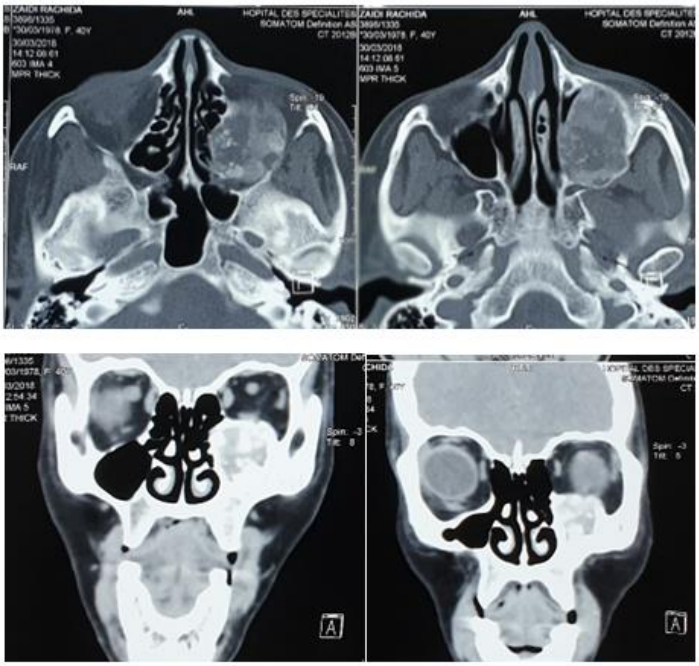

Fig-2: CT scan showing heterogeneous process of the left maxillary sinus, containing calcifications, and the infiltration of the left orbit floor.

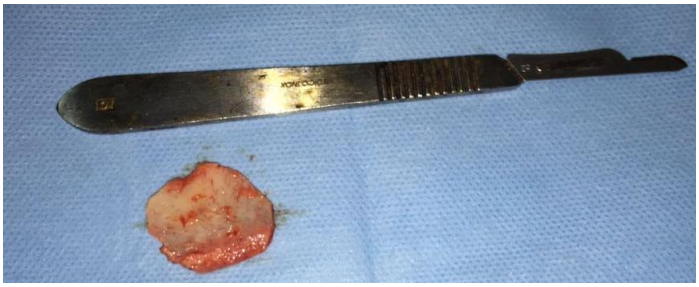

Fig-3: Graft of the iliac crest

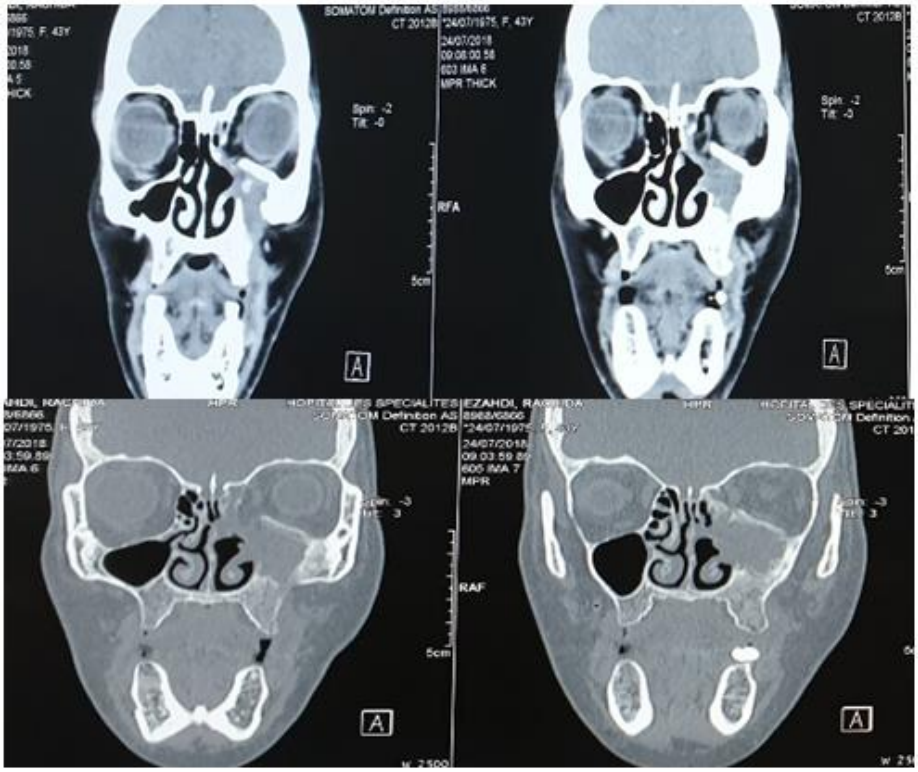

Fig-4: CT scan after 1year of surgery, showing the healing of the left maxillary sinus and the reconstruction of the floor with the iliac crest graft

Case N°2

A 45-year-old female patient, with diabetes on oral antidiabetics and already operated in 2007 for a left maxillary cyst with no documents of her first surgery. After 13 years of her first operation, the patient consulted for a left cheek swelling, that gradually increased in volume over a period of 04 months. Clinical examination found left vestibular expression of the tumor, with no other associated signs.

An orthopantomogram was performed that showed a clear radio image with well-defined edges, with root resorption of the adjacent teeth [fig.5]. The result of the orthopantomogram was insufficient to guide the diagnosis, hence the indication for a CT scan. That revealed a tissue process in the left maxillary sinus containing calcifications with a bone defect related to the 1st surgery [fig.6]. The mass measured 1.9cm by 3cm. The patient was operated, we did a vestibular incision, resected the tumor and then we performed a bone curettage.

Anatomopathological study confirmed the diagnosis of the maxillary ossifying fibroma. The postoperative follow-up was simple. No complications or recurrence on a follow-up of 02 years.

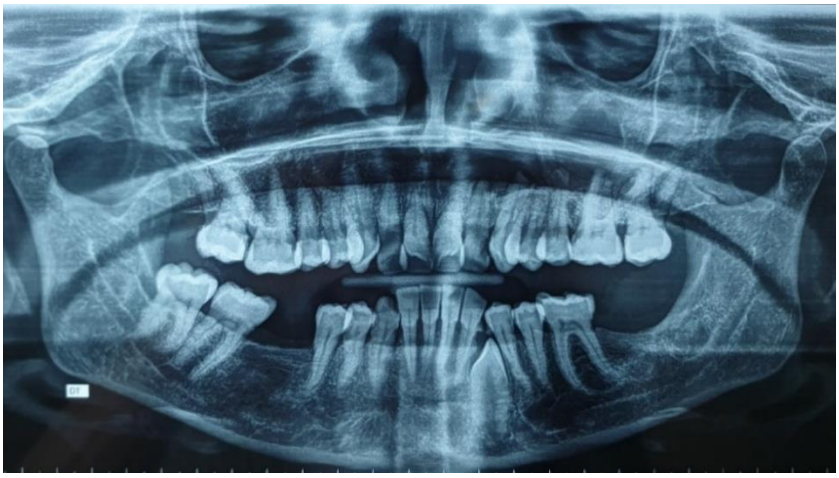

Fig-5: Orthopantomogram showing a clear radio, with root resorption of the adjacent teeth

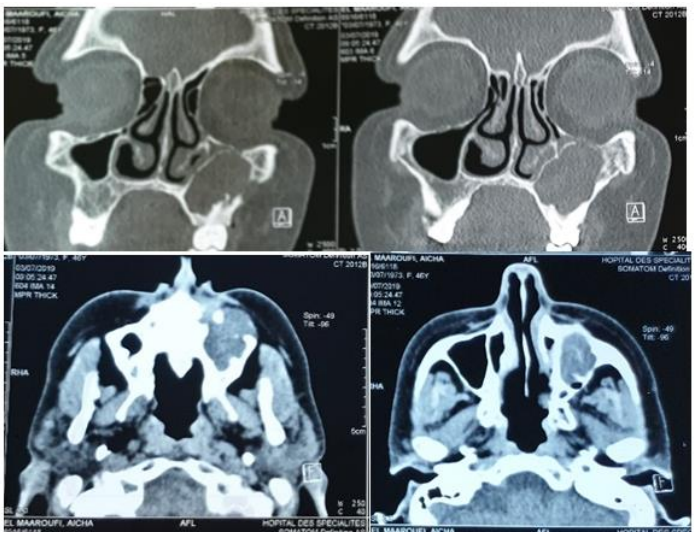

Fig-6: CT scan showing a tissue process in the left maxillary sinus containing calcifications with a bone defect related to the 1st surgery

DISCUSSION

Ossifying fibroma was included in the group of benign non-odontogenic tumors, according to the classification of the World Health Organization of 1992 [4].

The etiopathogenesis is still a subject of discussion, but the latest studies show that it originates from the mesenchymal cells of the periodontal ligament. These cells are able to form bone tissue as well as cement [5]. Currently, ossifying fibroma is classified as a benign odontogenic tumor of mesenchymal origin (WHO2017) [5].

Its evolution is insidious with a clinical polymorphism, but it remains of good prognosis. It occurs between the third and fourth decade of life [2]. Women are affected 2.5 times more frequently than men [3].

The maxilla is affected less than the mandible. The predilection sites are the molar and premolar regions for the mandible and the canine fossa for the maxilla [3].

Ossifying fibroma is asymptomatic in 31% of cases, and discovered incidentally during a routine radiological examination [3]. Clinically it manifests as a cheek swelling that is well limited, painless and slow growing [3].

The increase in the volume of the tumor can lead, depending on its location, mandibular or maxillary, to damaging the alveolo-dental nerve or the infraorbital nerve, to the displacement and root resorption of the adjacent teeth; and sometimes it can lead to the destruction of the maxillary sinus floor or the orbital frame [6, 8].

The initial radiological assessment is the orthopantomogram which can show us the location of the tumor, its density, give us an idea of the diagnosis and especially the evaluation of the state of the dental roots as well as the impact of the tumor on them [7]. But the results of this panoramic X-ray remain limited since the ossifying fibroma can have different aspects. It can present as a radiolucent lesion well limited with sometimes root resorption of the adjacent teeth; or as a multilocular image.

These different images can be confused with several lesions, especially fibrous dysplasia [7, 8]. Computed tomography has a large diagnosis ratio, according to the literature, the ossifying fibroma appears as a solitary radio transparent mass with lytic power, as well as the presence of punctate opacities occupying its center, they are called internal calcified components, minimal or absent at an early stage. At an advanced stage, the ossifying fibroma produces a very opaque, well-limited mass blowing the bone cortex [6]. In the study by Sciubba and Al [8], carried out on 18 cases of ossifying fibroma, the most frequent CT images were multi- or unilocular images with different degrees of opacity. Most of the tumors were well limited with root resorption of the adjacent teeth, without destruction of the other neighboring bone structures except for 2 cases where the tumor was very aggressive. The differential diagnosis is usually made at this stage with other lesions showing similar clinical signs and radiological images, especially fibrous dysplasia [9]. The well-defined radiographic appearance of the ossifying fibroma, the ease with which it can be separated from normal bone during surgery and especially the histological aspects are the main characteristics that differ it from fibrous dysplasia [10].

On the other hand, other diagnoses must also be taken into account as differential diagnoses [9, 10]: aneurysmal cyst, solitary bone cyst, giant cell

granuloma, calcifying odontogenic cysts, calcifying odontogenic tumors (Pindborg) and adenomatoid odontogenic tumors.

Anatomopathological study of the tumor remains the only way to confirm the diagnosis of ossifying fibroma. Histologically, ossifying fibroma is made up of fibrous tissue containing varied quantities of mineralized material such as bone or cement [4].

In our 2 cases the clinical and radiological characteristics were similar to what is reported in the literature. The 1st reported case underlines the destructive power that this tumor can have despite its benignity. The anatomopathological studies performed to confirm the diagnosis found consistent aspects with histological images of ossifying fibroma.

The treatment of ossifying fibroma is surgical, by enucleation and bone curettage for small tumors. For large tumors and when the surgical resection is extended, additional reconstruction with bone grafts and implants may be required [7].

CONCLUSION

Ossifying fibroma is a benign tumor; its diagnosis is based on the different clinical, radiological and histological evidence.

The management of ossifying fibroma is discussed case by case, it can range from simple excision, to the reconstruction of bone defects.

It is certainly a benign tumor, but the aggressive nature of it, which cannot be predicted, hence the importance of early management and complete excision of the tumor.

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