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Bimaxillary odonto-ameloblastic fibroma: A case report

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ABSTRACT

Introduction: Ameloblastic fibroma is a rare mixed odontogenic tumor, affects the young population, its management is mainly surgical. We report in this work the first observation of a concomitant bimaxillary localization.

Materials and Methods: This is a 31-year-old female patient with no pathological history who presented to our department for management of a maxillomandibular tumor. The clinical examination revealed a poor oral condition and a swelling of the alveolar ridges. The CT scan of the facial mass revealed a multilocular cystic lesion encompassing teeth in the maxillary and mandibular bone. The biopsy came back in favor of an odontoameloblastic fibroma. Management consisted of radical resection with reconstruction using local flaps. FOA is a tumor distinct from ameloblastoma, it affects the young patients without any predilection to gender. The radiological image is a mono or multilocular cystic image which poses a problem of differential diagnosis with other cystic tumors. The management is surgical, clinical and radiological postoperative surveillance is primordial given the risk of recurrence or sarcomatous transformation.

Conclusion: The FOA was for a long time considered as a form of ameloblastoma, is a rare tumor in the mandibular localization is the most frequent, the bimaxillary localization has never been described and the case we presented is the first in literature.

Keywords: Ameloblastic fibroma; Maxillomandibular; Radical surgery; Surveillance.

Introduction

Odonto-ameloblastic fibroma (OAF) or ameloblastic fibroma is a very rare mixed odontogenic tumor. It represents only 2% of odontogenic tumors. Histologically, the FOA is the result of an anarchic proliferation of the odontogenic epithelium. It usually evolves quietly, which delays its diagnosis, which in most cases is coincidental on routine oral clinical examination or on radiological examination. The tumor is localized and not very aggressive, hence the choice of conservative treatment as the reference treatment. Radical surgery may be indicated in case of extensive tumor, recurrence or malignant transformation. Ameloblastic fibroma often occurs in young subjects during the first two decades, without any gender-related predominance. The mandibular location is the most frequent. In this article, we present the case of a patient with a bimaxillary odonto-ameloblastic fibroma, discovered at the age of 31 years, which is rare.

Clinical Observation

The patient was 31 years old, married and mother of 3 children, with a history of trauma to the right mandibular

canine in childhood (at the age of 13 years), the patient had never been operated. The history of the disease goes back to the age of 14 years with the appearance of a small tumefaction in front of the 43, progressively and slowly increasing in volume. The patient consulted a dental surgeon who performed a resection of the tumefaction. The evolution was marked by recurrence after a few months, with the swelling becoming more extensive, involving almost the entire right mandibular hemiarcade, with concomitant appearance of similar mirror-like lesions on the right maxilla. The patient underwent several resections of the swollen gingiva with recurrence. The anatomopathological examination was inconclusive. The patient was then referred to our department for medical care.

The clinical examination of the face found a mandibular swelling slightly deforming the right labial commissure (Figure 1). On endobuccal examination, there was no limitation of the mouth opening, a deficient oral condition and the presence of smooth lobulated swellings on the right mandibular hemiarchade, covered with normal-looking gingiva and located on both sides of the alveolar ridge, encompassing the teeth. There is also a vestibular filling (Figure 2). In the maxilla, identical lesions to the mandibular lesions were present in the premolar-molar area (Figure 3). In view of this clinical situation, an orthopantomogram was requested (Figure 4), which showed an ectopic premolar tooth embedded in the basilar cortex in sector 4, with no bony abnormalities opposite the clinical lesions. A complementary CT scan was requested, which found multiple cystic images involving the alveolar ridge and respecting the basilar margin below and the sinus floor in the maxilla (Figure 5, 6).

A biopsy was scheduled in the dental chair under local anesthesia with excision of a significant fragment on the surface and in depth for histological analysis, which came back in favor of an odonto-ameloblastic fibroma. Management was surgical with resection of the entire mandibular alveolar ridge under the roots of the teeth while preserving the inferior alveolar nerve; the loss of substance was self-closing (Figure 7). In the maxillary region, the resection resulted in a larger loss of substance but without buccal-sinusal communication, which was filled with a BICHAT fat ball flap (Figure 8). Anatomopathological examination of the surgical piece confirmed the diagnosis. The evolution was marked by a good healing of the approaches with a metaplasia of the BICHAT ball after 7 days, then the patient was seen again at 3 then 6 months with a good evolution (Figure 9, 10).

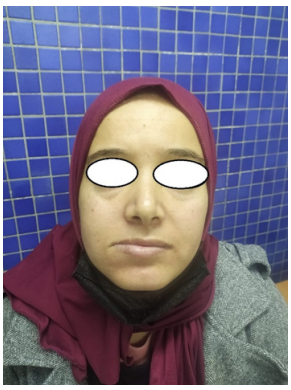


Figure 1. Patient with mandibular swelling with deformation of the right labial commissure.

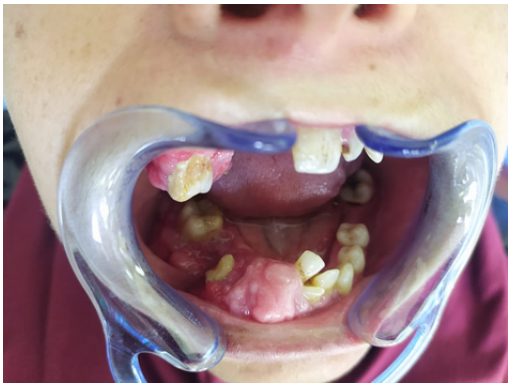


Figure 2. Clinical aspect of the mandibular lesion.

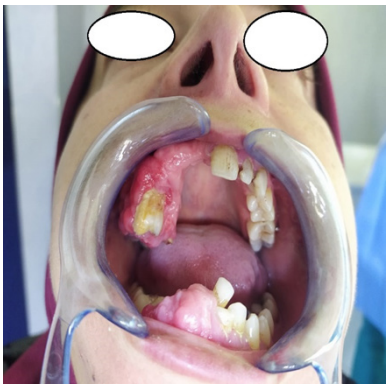


Figure 3. Aspect of the maxillary lesion

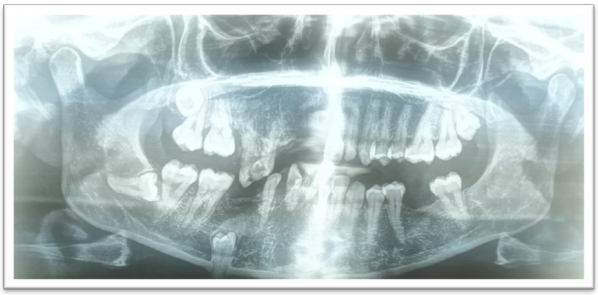


Figure 4. Orthopantomogram.

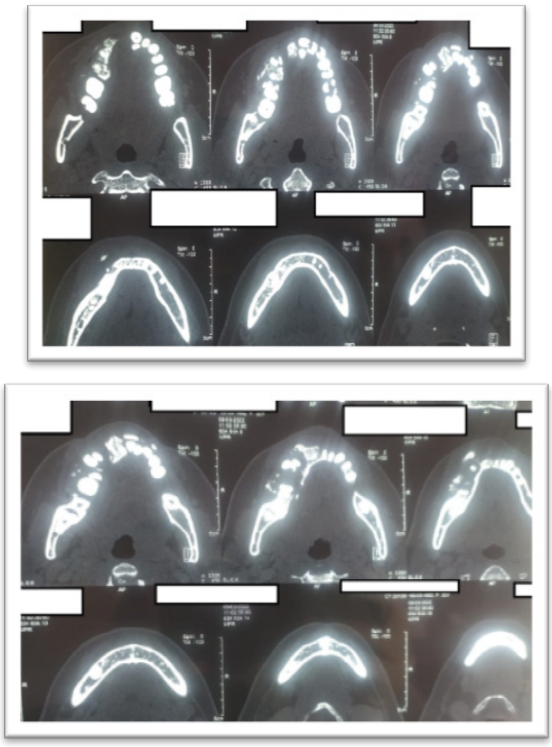


Figure 5. CT images of the tumor in axial section.

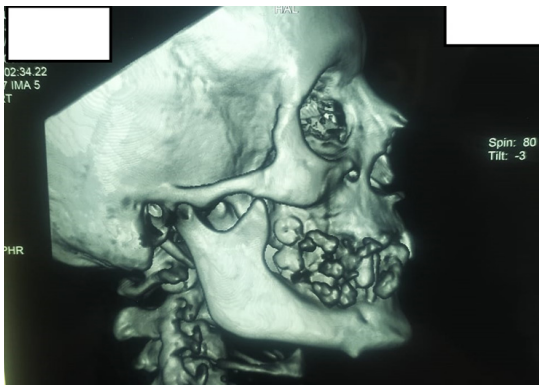


Figure 6. 3D reconstruction.

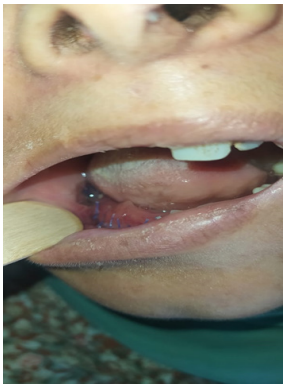


Figure 7. Postoperative aspect at the mandibular level.

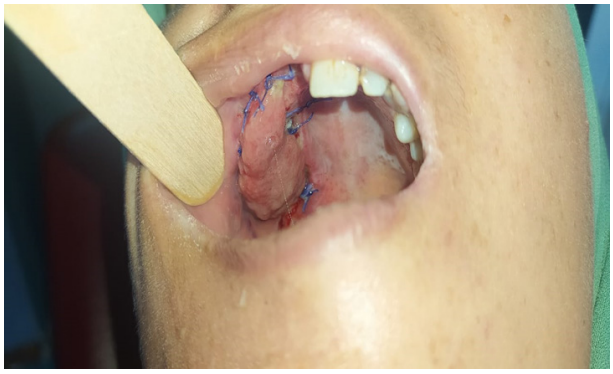
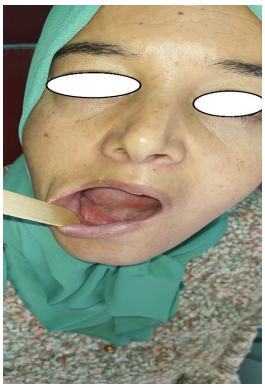


Figure 8. Post-operative appearance at the maxilla.



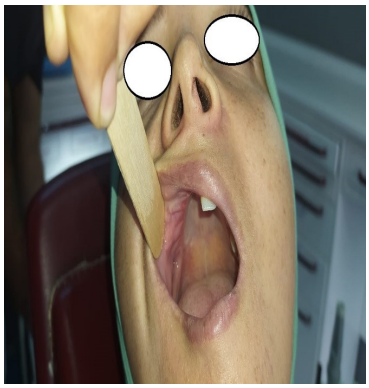


Figure 9. Clinical appearance at a distance.

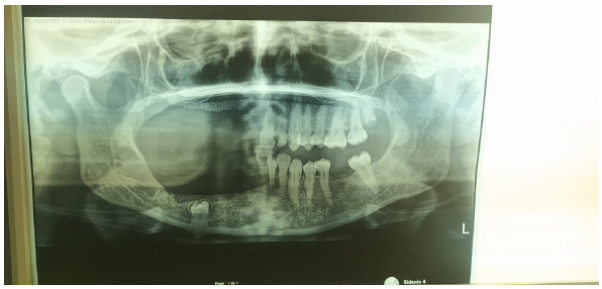


Figure 10. Control radiograph.

Discussion

Our observation is distinguished by the advanced age of the patient, the slow evolution of the tumor which is about 8 years and by the concomitant maxillo-mandibular localization of which no similar case has been found, to date, in the literature. Since 1891, FOA has been considered as a variant of ameloblastoma [1]. It was not until 1946 that Thoma and Goldman proposed to classify it as a separate entity due to its distinct characteristics from ameloblastoma [2]. The average age of onset is 15 years, without any gender predilection, in our patient the discovery was at the age of 31 years. Concerning the preferential location, the mandible is affected in 70% of cases, most often opposite an ectopic tooth as in our case. Because of the slow growth, the tumor may remain asymptomatic for a long time, which is a source of diagnostic delay. The diagnosis is then suspected fortuitously during a routine oral examination or in front of a dental radiography [1].

The radiological examination may show a well-limited, uni or plurilocular lacuna. The presence of a tooth within the tumor is found in 75% of cases. Other diagnoses may be evoked by the radiological aspect: a dental cyst, an odontogenic fibroma, an odontogenic keratocystic tumor or an ameloblastoma [3]. Anatomopathological examination reveals a double component with epithelial structures reminiscent of the enamel organ and areas of connective tissue simulating the dental papilla, without any calcified dental structure. These histological aspects may be confused with ameloblastoma. Sarcomatous transformation of the connective tissue is always possible and should be suspected by the presence of nuclear atypia, mitoses and loss of the epithelial component [4]. The FOA is generally not aggressive and has an excellent prognosis after enucleation [5]. Surgical treatment is the gold standard, but there is no consensus on whether or not radical removal is required. Enucleation of the tumor followed by curettage of the residual cavity is often proposed as a first step, whereas wide excision is recommended in the presence of large lesions extending to adjacent tissues and in case of recurrence, The question in these cases is whether or not to preserve the intra-tumoral tooth or teeth, and we believe that if the teeth do not interfere with the enucleation of the tumor, there is no reason to perform their extraction, leaving them the possibility of spontaneous eruption [6]. In our patient, a radical treatment was carried out with an exeresis of the tumor, taking away the teeth and the mucous membrane in front of it, considering the extent of the lesion, the included tooth was nevertheless preserved. The percentage of recurrence is very variable, it is around 70% after ten years, this percentage is lower after radical treatment than after conservative treatment. Recurrence of AFO is associated with inadequate surgical resection and may occur if tumor resection is not complete, especially for large tumors [7]. Regular surveillance is recommended to detect early recurrence or malignant transformation, the rate of which can be as high as 22% at 10 years. This risk of malignant transformation would increase if the patient's age at diagnosis was greater than 22 years [1].

Conclusion

Odonto-ameloblastic fibroma, formerly called ameloblastic fibroma, is a benign odontogenic tumor with atypical symptomatology whose diagnosis is based on a combination of clinical, radiological, and pathological arguments that distinguish it from ameloblastoma. The treatment of well-limited and small forms is enucleation with supported curettage; large AFOs can be treated with a conservative approach by enucleation and curettage, especially when the bony cortices are not blown and can be preserved. Radical treatment is indicated for soft tissue invasive lesions or recurrences.

Conflict of Interest

There is no conflict of interest to declare.

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