

Ameloblastic fibro-odontoma: a case report

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Summary

Ameloblastic fibro-odontoma: a case report.

The clinical case of an unusual ameloblastic fibro-odontoma (AFO) was reported. The patient's clinical chart as well as preoperative and postoperative radiographs and histological findings of a 20-year old man that addressed Dental Clinic at University of L'Aquila were thoroughly reviewed. The patient showed a swelling in the oral cavity and radiographic feature of a radiolucent lesion at left second premolar maxillary site. Histologic examination made diagnosis of AFO. AFO is a rare mixed odontogenic tumor with similarities to the ameloblastic fibroma (AF) and ameloblastic dentinoma. The nature and the relationships between mixed odontogenic tumors and related lesions are still controversial. Moreover is not clear if these lesions are separate pathologies or if they are different development stages of the same pathology.

Key words: ameloblastic fibro-odontoma (AFO), odontogenic tumor.

Introduction

The ameloblastic fibro-odontoma (AFO) is a rare, slow-growing, odontogenic tumor. This benign neoplasm has been defined by the World Health Organization (WHO) as "a neoplasm composed of proliferating odontogenic epithelium in a cellular ectomesenchymal tissue with varying degrees of inductive changes and dental hard tissue formation". The lesion has histologic feature and biologic behavior similar to the ameloblastic fibroma, but in

the AFO one or more cellular foci continue differentiation process and produce enamel and dentin.

This lesion is often an incidental radiographic finding. Radiographically, the tumor appear well circumscribed, round-to-ovoid radiolucency, surrounded by a thin sclerotic margin (1). According to the recent WHO classification of Odontogenic Tumors published in 2005, AFO is a benign tumor without invasive growth that belongs to the group of lesions with odontogenic epithelium with odontogenic ectomesenchyme, with or without hard tissue formation (2). There is considerable debate in literature regarding the relationship between AFO and other mixed odontogenic tumors. Some authors assert that AFO is a mature ameloblastic fibroma whereas other ones think it could be a precursor of odontoma (3).

It is rare in the jaw, where only about 2% of all cases have been reported. Focused literature revealed that neoplasm occur predominantly in children and young adults. An equivalent incidence in both upper and lower jaws and no gender predilection were reported (4).

Clinically, the size of the tumor shows marked variations, ranging from lesions detectable only microscopically, to giant tumors consisting of extensive calcified masses. Radiographs usually show a well-defined radiolucent area containing various amounts of radiopaque material of irregular size and form (2,5).

The aim of the current study was to report a clinical case of AFO and the long-term results after surgical treatment.

Case report

A 20-year-old Caucasian male was referred to the Dental Clinic at University of L'Aquila with an asymptomatic intra-oral swelling. Neither dental history reported local trauma or infection at lesion site, nor medical history revealed remarkable systemic diseases.

Panoramic radiography showed a rounded, well-defined, radiolucent lesion at upper left second bicuspid and first molar edentulous sites. It contained a radiopaque mass of apparently calcified material in proximity of the root of adjacent first bicuspid (Fig. 1). CT scan showed a 23x17mm osteolytic lesion in the left body of maxilla with lobulated and well-demarcated margins (Fig. 2). The borders of the lesion were in part radiopaque, similar to cortical bone (Fig. 3). Radiological findings were consistent with a benign bone tumour. Clinical examination revealed a circumscribed swelling in the vestibule on the left side of the maxilla, with an unaffected mucosa.

Under general anaesthesia, the tumour was removed through intra-oral approach. The first premolar was found to be involved by the mineralized mass.

Microscopically examination of sections stained with hematoxylin and eosin showed scattered cords of odontogenic epithelium surrounded by a large amount of cellu-



Figure 1 - Panoramic radiography.

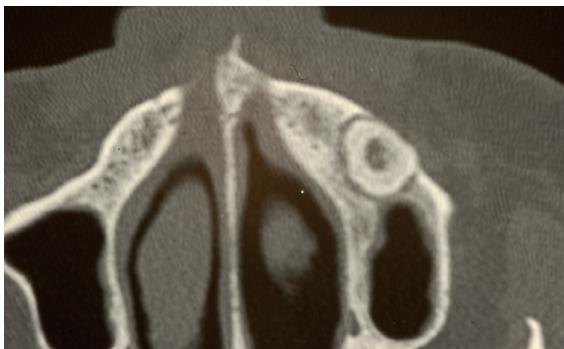


Figure 2 - CT – axial image.

lar connective tissue. In the mass of fibrous tissue, calcified areas were seen that were consistent with mature dentin formation and enamel matrix (3).

Post-operative clinical course was uncomplicated (Fig. 4). Finally, at 8 years-follow-up after implant-supported rehabilitation, no signs of recurrence were reported (Fig. 5).

Discussion

Histologic examination showed a benign ectomesenchymal neoplasia of odontogenic origin characterized by the proliferation of islands, nests and cords of epithelial cells. It exhibited a palisaded arrangement at the periphery and centrally a loose arrangement resembling the stellate reticulum of the enamel organ. The mesenchyme of the lesion was characterized by stellate or spindle-shaped cells, eosinophilic substance compatible with dentinoid material and basophilic material compatible with elementary enamel was observed (5).

The differential diagnosis of ameloblastic fibro-odontoma and ameloblastic fibroma is based on the presence or absence of elements indicative of differentiation of the tooth germ. The presence of both dentin and enamel is essential to call the tumor "Ameloblastic fibro-odontoma" (6). The distinction between developing complex odontoma and AFO is impossible sometimes. However the presence of great amounts of enamel, dentin, and cementum-like tissue arranged in a haphazard pattern suggests a diagnosis of odontoma (7).

Ameloblastic fibrodentinoma is considered by some authors as a stage between the ameloblastic fibroma (AF) and AFO based on extent of histodifferentiation (8,9). Many

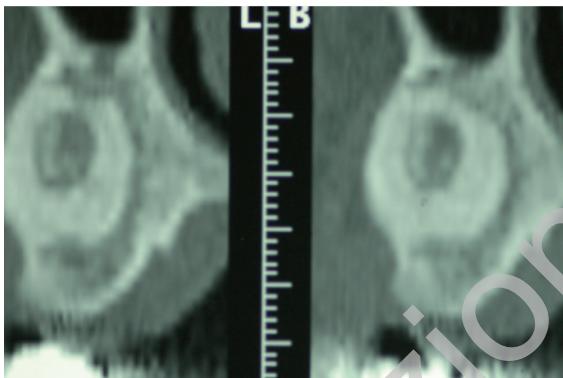


Figure 3 - CT – parasagittal image.

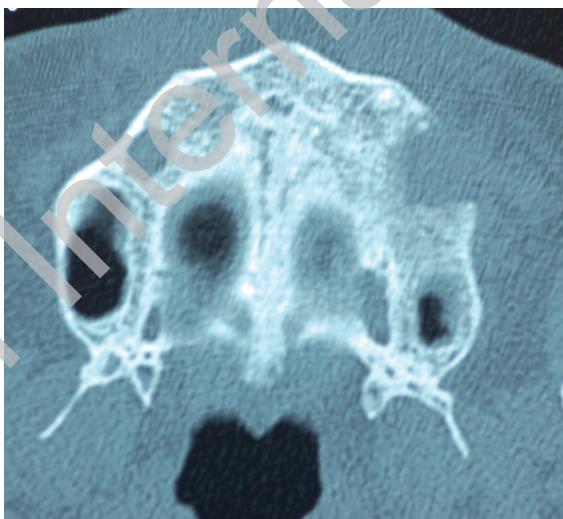


Figure 4 - CT – axial image after surgical treatment.

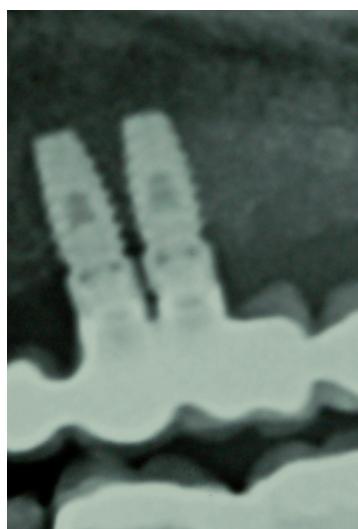


Figure 5 - Periapical radiography – 8 years-follow-up after implant-supported rehabilitation.

authors have reported that AFO is not aggressive and can be treated adequately with surgical curettage of the lesion without removing the adjacent teeth (10). The differential diagnosis for ameloblastic fibro-odontoma should include ameloblastic fibrosarcoma, a rare malignant counterpart to these odontogenic benign tumour, that arises in the jaws either de novo or from preexisting or recurrent ameloblastic fibroma. Finally, as AFO sometimes inhibits tooth eruption, the lesion can be microscopically differentiated from the follicular lesions around an impacted teeth in which a proliferation of odontogenic rests can occur. (11)

Conclusions

Treatment of AFO is generally enucleation. The associated tooth is normally removed, yet there are case reports of preservation of the involved teeth. Recurrence or local invasion is normally not observed if removed along with any involved teeth. Malignant transformation of ameloblastic fibromas has been rarely reported (12,13). The malignant transformation of an ameloblastic fibro-odontoma is even more rare (1,14-17). Potential transformation alone does not justify a radical treatment of all these benign lesions. As noted in the literature review, not all lesions previously classified as AFO are aggressive lesions, nor should they be expected to recur following conservative surgical intervention. When there is recurrence accompanied by changes in the histological pattern towards a more unorganized fibrous stroma with displacement of the epithelial component, more extensive treatment procedures are indicated (11).

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