

Ameloblastic fibro-odontoma in an unusual area of the maxilla: Two Case Reports.

Fibro-odontoma ameloblástico en una región inusual de los maxilares: reporte de dos casos.

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Receipt: 03/15/2019 **Revised:** 05/22/2019

Acceptance: 06/23/2019 **Online:** 12/30/2019

Abstract: Ameloblastic fibro-odontoma (AFO) is a mixed odontogenic tumor, which has an epithelial and mesenchymal component. It can be observed in imaging tests as a radiomixed lesion, with some authors claiming it represents only the predecessor of an odontoma. Epidemiologically, it appears between the second and third decade of life, predominantly in males and with a predilection for the posterior area of the mandible. This lesion presents a good prognosis and it responds well to enucleation by curettage with a low rate of recurrence. In the present article, two cases of ameloblastic fibro-odontoma in an unusual region of the maxilla are presented.

Keywords: *Ameloblastic fibro-odontoma; odontoma; odontogenic tumors; maxilla; tooth abnormalities; adolescent.*

Resumen: El Fibro-odontoma ameloblástico (FOA), es una lesión tumoral mixta de origen odontogénico, el cual tiene un componente epitelial y mesenquimático. En su imagenología se observa como una lesión radiomixta, debido a que algunos autores afirman que este no es más que el predecesor de un odontoma. Epidemiológicamente, se presenta entre la segunda y tercera década de la vida, preferencia por sexo masculino y predilección por el área posterior de la mandíbula. Es una lesión de buen pronóstico que responde bien a la enucleación por curetaje con baja tasa de recidiva. En el presente artículo, se presentan dos casos de fibro-odontoma ameloblástico en una región inusual de los maxilares.

Palabras Clave: *Fibro-odontoma ameloblástico; odontoma; tumores odontogénicos; maxilar; anomalías dentarias; adolescente.*

INTRODUCTION.

AFO is a benign tumor with a mixed origin that has a mesenchymal and epithelial component, forming mineralized tissues and is less common than ameloblastic fibroma (AF).^{1,2} Both the odontoma and the AFO present similar characteristics³ and some authors refer that the AF only represents the predecessor stage of an odontoma⁴ while others like Ruiz *et al.*,⁵ disagree, affirming that the AFO is a AF variant which occurs together with an odontoma, since the latter is completely radiolucent in the imaging.

The AFO appears between the first and second decade of life, mainly affecting males between 9 to 15 years of age. Some authors state that the most common site for this type of lesions is mainly the posterior region of the mandible (52% - 62%) associated with an impacted tooth.⁶⁻⁸ Its histologic structure consists

Cite as:

Valencia D, Jiménez E & Urbano S. Ameloblastic fibro-odontoma in an unusual area of the maxilla: Two Case Reports. J Oral Res 8(6):517-521.

Doi:10.17126/joralres.2019.075

of thin strands and cords of cubic odontogenic epithelial cells, embryonic fibrous connective tissue and primitive odontogenic epithelium, simulating the stellate reticulum which rests on embryonic connective tissue containing randomly oriented fibroblasts, widely spaced.⁹

As an encapsulated lesion, AFO is non-invasive and easily removed in most cases. The treatment consists of conservative surgery that can be enucleation by curettage; however, for large lesions, bloc resection is suggested to avoid recurrence.¹⁰ The aim of the present clinical cases is to inform that the imaging characteristics of certain sites of the maxilla should not be considered highly conclusive for a diagnosis, despite the fact that it can be compared to any other type of pathology according to imaging patterns and epidemiological features.

CASE REPORT 1

14 year-old boy presented to our consultation on an orthodontic referral due to a slight increase in volume with approximately 13 months of evolution in the left canine site with persistence of deciduous teeth and absence of permanent ipsilateral canine being asymptomatic.

Upon X-ray examination, a radiomixed lesion of approximately 13.5mm in diameter was found in the upper left canine site with the impaction of a permanent canine with preservation of the cortex and without radicular displacement or reabsorption of adjacent teeth. Upon tomographic imaging, an impacted canine with a

hypodense pericoronary space was observed, suggesting a dentigerous cyst as a diagnostic impression and well-defined hyperdense multilocular foci, associated with a composite odontoma, contemplating a hybrid lesion (Figure 1A).

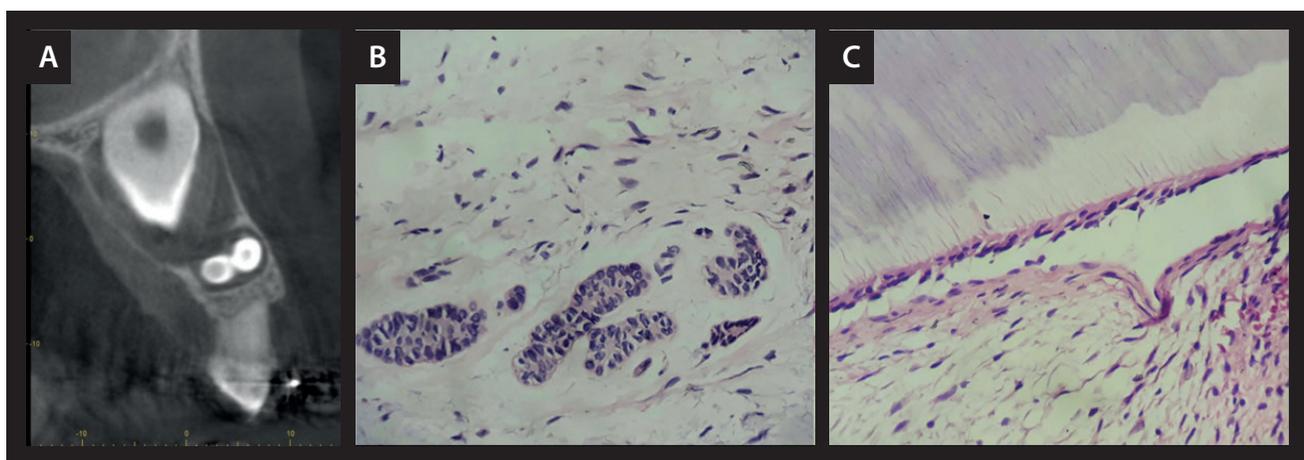
In a surgical meeting consisting of oral surgery and orthodontics, we decided to perform enucleation of the lesion and allow a spontaneous canine eruption due to incomplete development of the root; otherwise, canine traction would be performed posteriorly. Once the patient signed an informed consent form, complete enucleation of the lesion was performed under local anesthesia, revealing a soft and brown consistency.

A sample was sent for histopathological analysis with diagnostic impression of dentin cyst, ameloblastic fibroma, composite odontoma and complex odontoma.

The pathology report shows that there is a lesion consisting of islands of inactive odontogenic epithelium surrounded by connective tissue in myxoid areas. Areas of mineralized tissue reminiscent of cementum are identified, in addition to dental structures, enamel, dentin and pulp tissue (Figure 1B y Figure 1C).

At the 6-month post-surgical check-up, circum-coronary bone neoformation with eruption in an oblique direction was evident, and it was determined that orthodontics should be performed, leaving adjacent lateral teeth free to avoid reabsorption, and canine traction with orthodontic button.

Figure 1: Tomographic image and pathology report.



A: Sagittal section showing impaction of the upper left permanent canine and denticles. **B:** Histological section where cords and epithelial islands are observed. **C:** Histology with dental structures and components.

CASE REPORT 2

12 year old female patient attending oral surgery consultation for delayed eruption in canine, lateral and central upper right region, with persistence of canine and lateral deciduous ipsilateral. During clinical examination palpation of a tumor lesion of stony consistency located in the upper right dental arch occurred, with normochromic mucosa covering it, with respect to the adjacent mucosa, asymptomatic, approximately 2cm in diameter, of unknown evolution time and etiology, whose presumptive diagnosis was odontoma.

Tomographically, a hypodense unilateral image of approximately 25mm x 20mm was evident, in which lateral and central upper right canines, at Nolla 9 stage, are included; in addition, multiple hyperdense images

of varied dimensions are observed, with the appearance of denticles and amorphous content, with the presence of lateral supernumerary (Figure 2A, Figure 2B and Figure 2C). After signing informed consent by the legal representative, and under local anesthesia, we proceed to perform exodontia of deciduous teeth and enucleation of the content of the lesion, of dark brown color and soft consistency about 0.3cm, which was deposited in 10% formaldehyde for anatomopathological study with diagnostic impressions of complex odontoma, cystic odontogenic tumor and adenomatoid odontogenic tumor.

The histopathological report informs a benign mesenchymal lesion composed of a proliferation of spindle cells with tapered ends, resting on a lax matrix with myxoid changes.

Figure 2: Sagittal, transversal and coronal sections, where the inclusion of central and upper right permanent incisors, denticles and hyperdense lesion are observed.

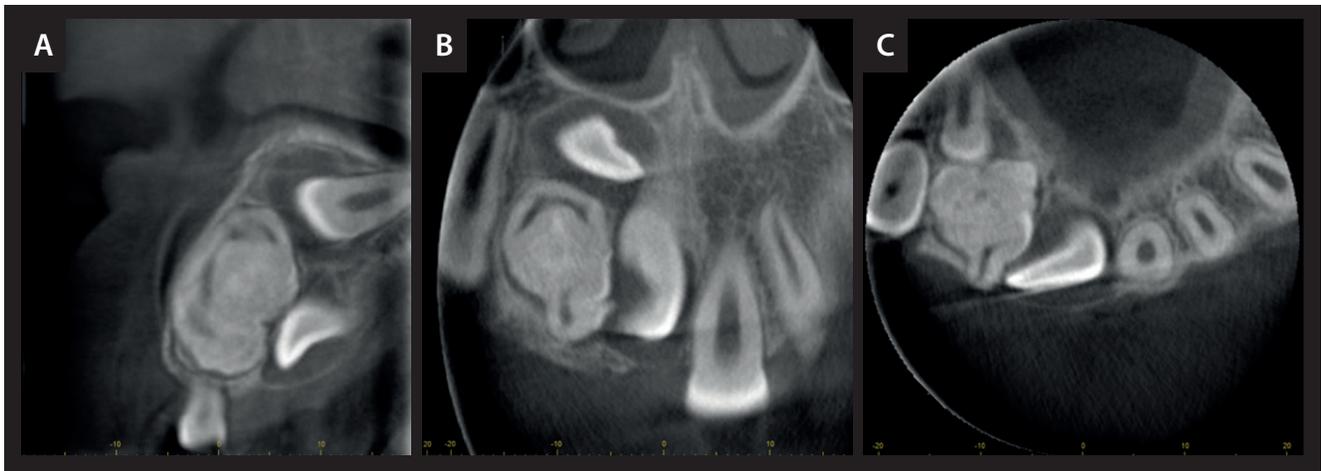
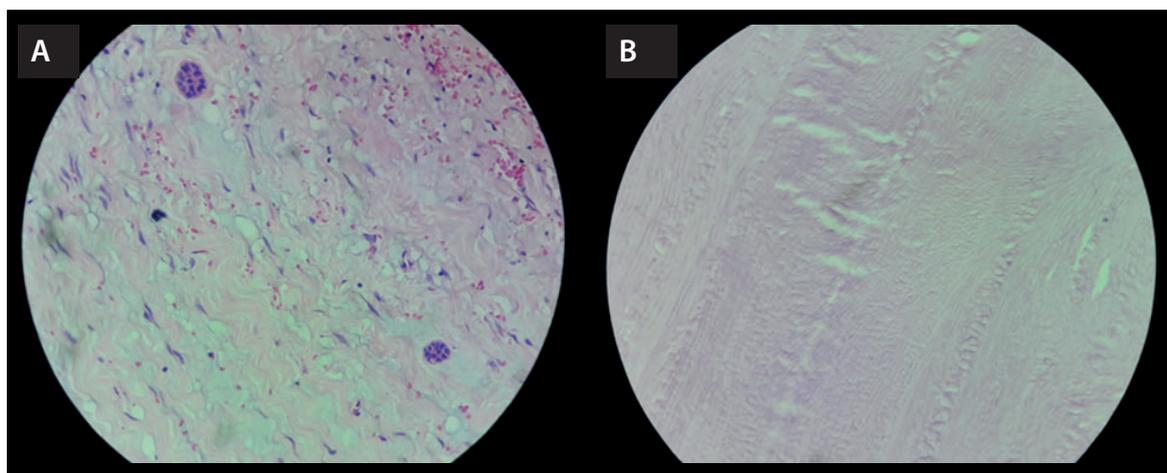
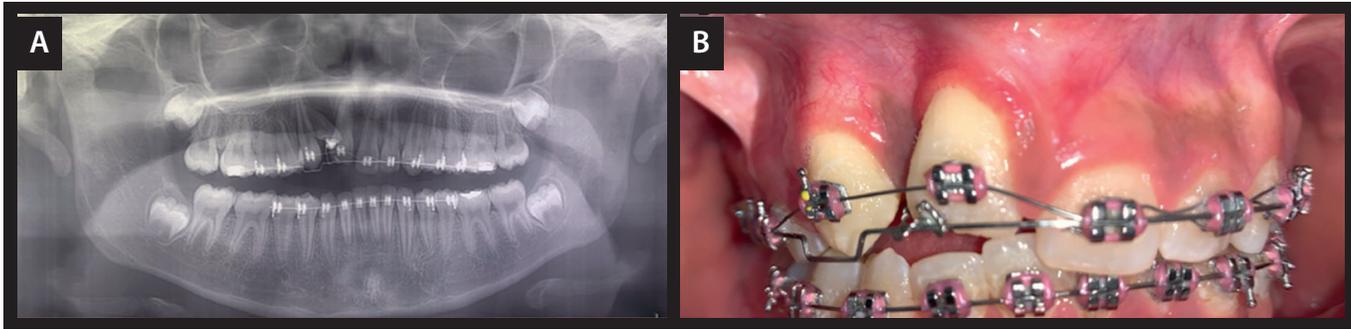


Figure 3: Histopathological report .



A: Microphotography 10x. Mesenchymal lesion with small areas of ameloblastic epithelium, resting on a fibromyxoid stroma. **B:** Photomicrograph 40x. Enamel matrix adjacent to the lesion.

Figure 4: Panoramic radiograph with orthodontic appliances.



A: Panoramic radiograph with orthodontic appliances positioned for traction of central incisor, lateral and upper right canine. **B:** Central and canine with orthodontic appliances for traction to their desired position.

Small light tubular structures are recognized, covered by a double layer of cubic cells without cytological atypia. The lesion is covered by a stratified cubic epithelium of usual morphology, without evidence of necrosis foci or mitosis (Figure 3A and Figure 3B)

At one month follow-up, the evolution showed coronary bone neoformation. The patient was followed-up at 8 months, when central, lateral and upper right canine incisors are observed, which are in traction to the adequate position (Figure 4A and Figure 4B).

DISCUSSION.

The AFO is a mixed odontogenic tumor of rare appearance, which is accompanied by structures similar to teeth. It can interrupt tooth eruption or displace affected teeth, but the teeth involved remain vital without root resorption.¹¹ Radiographically, a well-defined radiolucent area is evident, which contains a variable amount of radiopaque material. The histopathological component confirms epithelial cords within a primitive cellular ectomesenchyme that simulates dental papilla, dentin and enamel.¹²

In a study of 108 lesions of this type, Kirjavainen *et al.*,¹⁴ reported that they occurred between the ages of 6.3 and 9.6 years with an incidence rate of 1:1.65 in female to male patients, up to age 20, with a predilection for the posterior mandibular area in the molar region, associated with an unerupted tooth in 94% of cases, a recurrence rate of 6% and a good prognosis.^{13,15}

On the other hand, Sardana *et al.*,¹⁶ state that the incidence is practically the same for both jaws, with greater frequency in the posterior region.

Nayak *et al.*,¹⁷ and Alani *et al.*,¹⁸ reported one case of AFO in the anterior region of the maxilla, and one with involvement of the maxillary sinus, respectively, with retained teeth and intra- and extra-oral volume changes, highlighting the similarity to the cases presented here.

CONCLUSION.

The development process of dental organs has a great potential to lead to pathologies in any region of the jaws regardless of the site of predilection of certain neoplasms, therefore, other pathological processes should not be ruled out despite a low incidence rates in certain sites. It is important to take into account the chronology of dental eruption in relation to the age of the patients, in relation to the appearance of new tumor lesions associated with retained teeth, as in the present cases.

Conflict of interests: The authors declare no conflict of interest of any kind.

Ethics approval: Patients gave informed consent.

Funding: Self-funded

Author's contribution: All authors contributed to the manuscript.

Acknowledgements: None.

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