Casos Clínicos
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Ameloblastic fibro odontoma; clinical case report

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Abstract: Ameloblastic fibro-odontoma is an infrequent benign odontogenic tumor with cubic odontogenic epithelia cells, embryonic fibrous connective tissue, and primitive odontogenic epithelium. It mainly affects children and young adults between the first and second decade of life. This article is a clinical case report of a 12 - year - old male patient seen at Hospital of Pediatric Dentistry "Don Benito Quinquela Martín," where he was diagnosed with a benign odontogenic tumor associated with an impacted permanent lower right canine tooth. The treatment plan included a multidisciplinary collaboration among the Departments of Preventive Dentistry, Surgery, Radiology, and Orthodontics. In the first stage, prevalent infections were treated and the host was strengthened. The second stage was the surgica removal of the lesion and associated dental structures. The patient's course was favorable and continued his treatment at the Department of Orthodontics. **Objective:** To describe the clinical course of a 12-year-old male patient diagnosed with ameloblastic fibro-odontoma managed with an interdisciplinary approach in the setting of a comprehensive treatment plan. **Conclusion:** An interdisciplinary management in the setting of a comprehensive plan resulted in a favorable approach to ameloblastic fibro-odontoma.

Key words: Early diagnosis, ameloblastic fibro odontoma, surgery, impacted tooth

Fibro - odontoma ameloblástico; relato de caso clínico

Resumo: O fibro - odontoma ameloblástico é um tumor odontogênico benigno com células epiteliais odontogênicas cuboides, tecido conjuntivo fibroso embrionário e epitélio odontogênico primitivo. Se apresenta com pouca frequência e afeta principalmente crianças e adultos jovens entre a primeira e segunda década de vida. Este trabalho é um relato de caso clínico de um paciente de sexo masculino de 12 anos de idade atendido no Hospital de Odontologia Infantil "Don Benito Quinquela Martín"; diagnosticado com um tumor odontogênico benigno associado a um canino permanente inferior direito impactado. É realizado um plano de tratamento integral, individualizado e com forte componente preventivo, trabalhando sempre em constante inter-relação entre os Serviços de Odontologia Preventiva, Cirurgia, Radiologia e Ortodontia do hospital. Numa primeira fase, são tratadas as infecções prevalentes e é realizado o reforço do hospedeiro. Posteriormente é realizada a intervenção cirúrgica para remover a lesão e as estruturas dentárias associadas a ela. A evolução do paciente foi favorável, permitindo que ele continue o tratamento no Serviço de Ortodontia. **Objetivo:** Descrever a situação clínica de um paciente masculino de 12 anos de idade, com diagnóstico de fibro - odontoma ameloblástico com uma abordagem interdisciplinar e dentro de um plano de tratamento integral. **Conclusão:** O tratamento realizado com uma abordagem interdisciplinar e dentro de um plano integral, permitiu alcançar um resultado favorável na abordagem do fibro - odontoma ameloblástico.

Palabra-chave: Diagnóstico precoce, fibro - odontoma ameloblástico, cirurgia, dente impactado.

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Fibro - odontoma ameloblástico; reporte de caso clínico

Resumen: El fibro - odontoma ameloblástico es un tumor odontogénico benigno con células epiteliales odontogénicas cúbicas, tejido conjuntivo fibroso embrionario y epitelio odontogénico primitivo. Se presenta con poca frecuencia y afecta principalmente a niños y adultos jóvenes entre la primera y segunda década de vida. Este trabajo es un reporte del caso clínico de un paciente de sexo masculino de 12 años de edad atendido en el Hospital de Odontología Infantil "Don Benito Quinquela Martín", al cual se le diagnostica un tumor odontogénico benigno asociado a un canino permanente inferior derecho impactado. Se realiza un plan de tratamiento integral, individualizado y con fuerte componente preventivo, trabajando siempre en constante interrelación con los servicios de Odontología Preventiva, Cirugía, Radiología y Ortodoncia del hospital. En una primera etapa se tratan las infecciones prevalentes y se realiza el refuerzo del huésped. Posteriormente se realiza la intervención quirúrgica para extirpar la lesión y las estructuras dentarias asociadas a esta. La evolución del paciente fue favorable permitiendo continuar su tratamiento en el servicio de Ortodoncia. **Objetivo:** Describir la situación clínica de un paciente masculino de 12 años de edad, con diagnóstico de fibro - odontoma ameloblástico con un enfoque interdisciplinario y dentro de un plan de tratamiento integral. **Conclusión:** El tratamiento realizado con un enfoque interdisciplinario y dentro de un plan integral, permitió lograr un resultado favorable en el abordaje del fibro - odontoma ameloblástico.

Palabras clave: Diagnóstico precoz, fibroodontoma ameloblástico, cirugía, diente impactado.

Introduction

According to the World Health Organization Classification of Tumors, published in 2005, ameloblastic fibro-odontoma (AFO) is part of the group of lesions with odontogenic epithelium containing odontogenic ectomesenchyme, with or without tissue formation (Table 1).[1]

Table 1: *Histological classification of benign odontogenic tumors (WHO, 2005)*

1. Odontogenic epithelium with mature fibrous stroma, without odontogenic ectomesenchyme	**2. Odontogenic epithelium with odontogenic ectomesenchyme, with or without tissue formation**
Ameloblastoma, solid/multicystic type	Ameloblastic fibroma
Ameloblastoma, extraosseous/peripheral type	Ameloblastic fibrodentinoma
Ameloblastoma, desmoplastic type	Ameloblastic fibro-odontoma
Ameloblastoma, unicystic type	Odontoma
Squamous odontogenic tumor	Odontoma, complex type
Adenomatoid odontogenic tumor	Odontoma, compound type
Keratocystic odontogenic tumor	Odontoameloblastoma
Tumor odontogênico queratocístico	Calcifying cystic odontogenic tumor
	Dentinogenic ghost cell tumor
3. Mesenchyme and/or odontogenic ectomesenchyme, with or without odontogenic epithelium	**4. Bone related lesions**
Odontogenic fibroma	Ossifying fibroma
Odontogenic myxoma (myxofibroma)	Fibrous dysplasia
Cementoblastoma	Osseous dysplasias
	Central giant cell lesion (granuloma)
	Cherubism
	Aneurysmal bone cyst
	Simple bone cyst

AFO is similar to ameloblastic fibroma, which shows inductive changes that allow the formation of varying degrees of enamel and dentin matrix.[1] Some authors state that ameloblastic fibroma and odontoma, complex type, may be combined in this lesion.[2]

AFO is an infrequent benign odontogenic tumor.[3] The etiology of AFO is still unknown and considered controversial because it mimics the ontogeny of development of tooth germs during the embryonic stage, although in an aberrant manner, evidencing different degrees of inductive interactions between the epithelium and the odontogenic ectomesenchyme, which translate into the formation of enamel and dentin inside the mesodermal cell tissue.[4]

It is a well-circumscribed, slow-growing lesion usually located in the maxillary or mandibular posterior region that causes cortical expansion and tends to increase volume. Most cases are associated with unerupted teeth.[5,6] In general, AFO is asymptomatic, so the detection of this type of lesions takes place during routine X-ray reviews.[7] AFO mainly affects children and young adults between the first and second decade of life, at a mean age of 14 years.[8] No sex prevalence has been observed, although some authors state that there is a mild tendency towards males.[9]

Radiographically, AFO exhibits a well-defined unilocular or multilocular radiolucent appearance, with varying levels of radiopacity inside, similar to dental tissue in different stages of odontogenesis. In addition, it may cause cortical plate expansion, so it is difficult to differentiate it from other lesions of similar appearance, such as ameloblastoma or dentigerous cyst if associated with an unerupted tooth.[10]

Histologically, AFO is an encapsulated lesion made up of thin cords and strands of cubic odontogenic epithelial cells, embryonic fibrous connective tissue, and primitive odontogenic epithelium, mimicking the stellate reticulum.[11]

AFO treatment is associated with a conservative surgical approach. The sporadic recurrence of AFO has been attributed to an inadequate surgical extraction in the beginning of treatment. According to some other reports, a conservative enucleation is enough.[12] The bibliography describes a controversy about the extraction or retention of the associated tooth germ in the case of AFO. Most articles suggest the extraction of the associated tooth germ to prevent recurrence.[6, 13,14]

The objective of this report is to describe the clinical case of a 12-year-old male patient diagnosed with ameloblastic fibro-odontoma managed with an interdisciplinary approach in the setting of a comprehensive treatment plan.

Case report

This was a 12-year-old male patient with no personal or family medical history of interest, who attended Hospital of Pediatric Dentistry “Don Benito Quinquela Martín” seeking care; the reason for consultation was “cavities”. The patient had previous experience with a dentist in a different facility. He was admitted for care at the Department of Preventive Dentistry.

The intraoral clinical examination showed persistence of the lower right primary canine tooth (8.3), absence of the lower right permanent canine tooth (4.3), and convexity of the vestibular bone plate in the area of said canine tooth, without spontaneous pain or tenderness (Figures 1 and 2).

Figure 1. *Intraoral clinical appearance. Persistence of tooth 8.3 (lower right primary canine).*

Figure 2. *Convexity of the vestibular bone plate in the area of tooth 4.3 (lower right permanent canine).*

The radiographic examination included panoramic and periapical X-rays (Figures 3 and 4), which showed that tooth 4.3 was retained and its crown was surrounded by a radiolucent image of more than 2 mm in diameter and presence of radiopaque images compatible with denticles.

Figure 3. *Preoperative panoramic dental X-ray. Persistence of tooth 8.3 (lower right primary canine), image compatible with odontoma and retained tooth 4.3 (lower right permanent canine).*

Figure 4. *Preoperative periapical X-ray.*

A comprehensive, individualized treatment plan with a strong preventive component was implemented, including preventive and therapeutic actions based on individual risk to make an adequate diagnosis. The comprehensive assessment should include general condition and growth, extraoral and intraoral soft tissue, gingival and periodontal health and hygiene, occlusion development, and child behavior.[15]

Once the corresponding explanations were provided to the patient's parents, they signed the informed consent form established by the Government of the Autonomous City of Buenos Aires - Ministry of Health (Resolution 0356/ MSGC/09 - Law 153 - Decrees 208/01 and 2316/03) for the performance of the proposed treatment and for the registration, documentation, and potential publication of the case report.

While the comprehensive, individualized treatment plan with a strong preventive component was implemented, the Department of Pediatric Dentistry consulted with the Departments of Surgery and Orthodontics of the hospital.

The Department of Surgery completed the X-ray examination using Clark's rule (Figure 5). The X-ray showed that with cone distalization, mesial displacement of the lesion in the opposite direction occurs, so it is considered to be adequate for surgical approach through the vestibular area.

Together with the Department of Orthodontics, the occlusion was studied and tooth 4.3 (lower right permanent canine) was deemed to be in an unfavorable position, with potential lesion recurrence. Tooth 8.3 (lower right primary canine) and tooth 4.3 (lower right permanent canine), which was retained, were extracted and the lesion was removed.

The preoperative visit was completed with clinical laboratory analysis. Considering that the patient had previous experience at the Department of Surgery of the hospital, where he had undergone the extraction of tooth 2.6 (upper left first molar) and tooth 3.6 (lower left first molar), motivation was

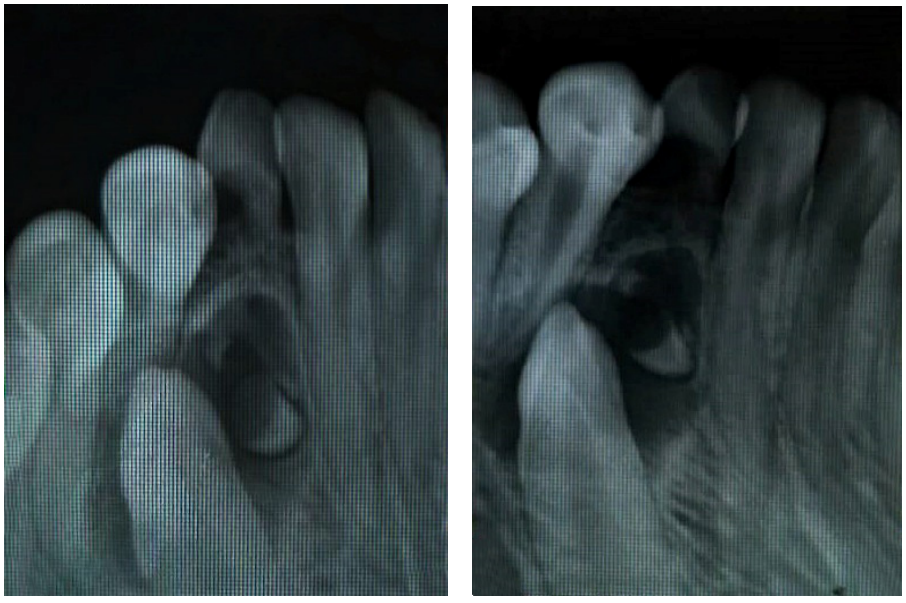

Figure 5. *Clark's rule.*

reinforced and the relevant explanations about the procedure were provided to the family, so as to favor their collaboration and the success of the surgery.

Surgical technique

Intraoral antisepsis was done with 0.12% chlorhexidine and, using local nerve block with 4% articaine hydrochloride 4% - epinephrine at a 1:100 000 ratio to raise the mucoperiosteal flap. The lower right primary canine tooth (8.3) and the retained lower right permanent canine tooth (4.3) were extracted using the tooth sectioning technique as surgical resource (Figures 6 and 7). A lower incisor forceps, a straight elevator, and a cylindrical turbine burr were used.

The cystic pericoronary sac of tooth 4.3 (lower right permanent canine) and the denticles observed in the X-ray were removed, and the procedure was completed by taking an intraoperative periapical X-ray to corroborate total extraction; the incision was sutured using synthetic, non-resorbable thread (Figures 8A, 8B, and 8C).[8] The patient's prescriptions included antibiotics (amoxicillin-500 mg) and analgesics (ibuprofen-400 mg) orally and mouthwash with chlorhexidine 0.12% at night, once daily, for a week.

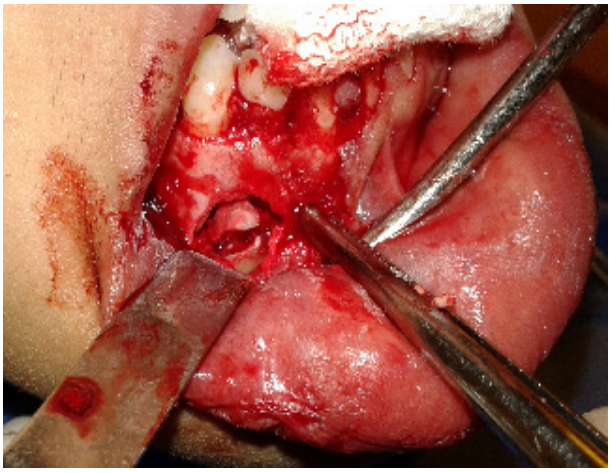

Figure 7. *Sectioning of tooth 4.3 (lower right permanent canine).*

The specimen of hard tissue (bone and tooth) and soft tissue (membrane) were sent, according to protocol, to the Laboratory of Surgical Pathology of the School of Dentistry of Universidad de Buenos Aires (Figure 9).

Figure 6. *Intraoperative image. Retained tooth 4.3 (lower right permanent canine) and ameloblastic fibro-odontoma.*

Figure 8A. *Surgical site.*

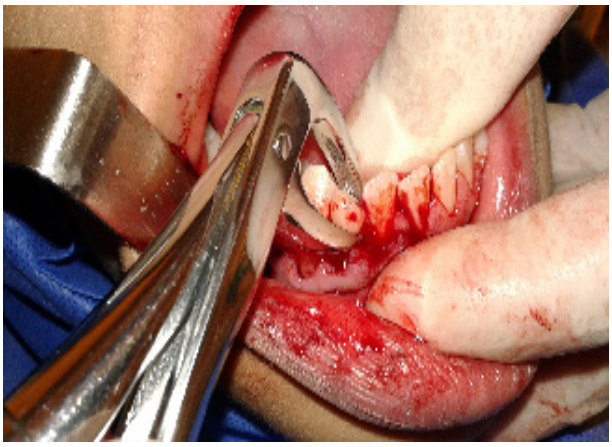

Figure 8B. *Extraction of tooth 8.3 (lower right primary canine).*

Figure 8C. *Suture.*

The first postoperative control was done at 7 days. Suture stitches were removed, and the patient was referred to the Department of Endodontics for a sensitivity test of tooth 4.2 (lower right lateral incisor) given its proximity to the lesion. The test was positive for sensitivity.

The pathology study reported the following: "Histopathological finding corresponding to an odontoma-like odontogenic tumor compatible with ameloblastic fibro-odontoma."

Figure 9. *Tooth 4.3 (lower right permanent canine) with tooth sectioning and removed denticles.*

Postoperative clinical radiographic controls were done at 1, 3, and 6 months. Controls showed normal mucous membranes, with no associated diseases and bone regeneration according to the postoperative period.

Based on the treatment plan implemented, the patient continues receiving care at the Department of Preventive Dentistry and has been admitted for treatment to the Department of Orthodontics, and all departments involved continue working in an interdisciplinary manner (Figures 10 and 11).

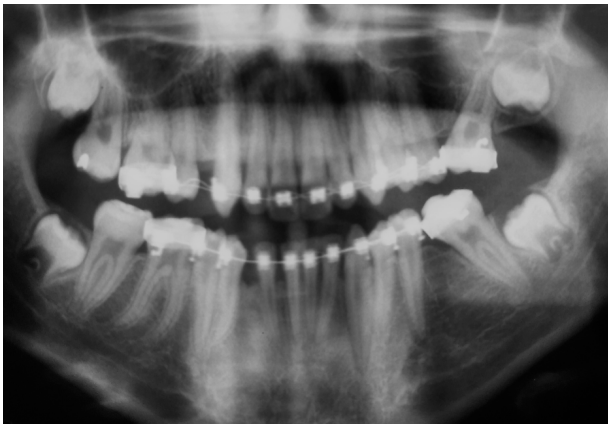

Figure 10. *Postoperative panoramic dental X-ray. Control at 2 years.*

Figure 11. *Current intraoral appearance.*

Discussion

In 2005, the WHO established that AFO is a benign neoplasm consisting of proliferative odontogenic epithelium, included in an odontogenic ectomesenchyme resembling the dental papilla, with varying degrees of inductive changes and formation of dental hard tissue.[1,7] Other authors have directly associated AFO with an immature stage of odontoma, complex type.[7,16]

Notwithstanding the above, AFO should not be considered an odontoma, because there have been case reports showing neoplastic behaviors, such as recurrence and malignant change, even after a careful lesion excision, including displaced unerupted teeth.[17]

In this case, the lesion was diagnosed due to the clinical persistence of the lower right primary canine tooth, complemented by a periapical X-ray of the area, which allowed to note the retention of tooth 4.3 (lower right permanent canine) and the presence of AFO. This is a confirmation of the importance of taking an X-ray before performing any tooth extraction.

A complementary radiographic study in this clinical case was the use of Clark's rule, because, at that time, our hospital did not have a CT scanner to check the location of the retained tooth to define the area for surgical approach. At present, the potential referral to another public hospital for a CT scan allows us to establish the certain location of teeth to be treated and their relation to adjacent tissues.

According to some studies, AFO may show an aggressive behavior with multiple recurrences.[18,19] It has been suggested that a mainly benign case of AFO may undergo malignant transformation into ameloblastic fibrosarcoma. Therefore, it is important to continue with AFO follow-up for several years to ensure that no aggressive recurrence occurs in the future.[20]

This patient is still receiving follow-up with clinical and radiographic controls. Findings of bone neoformation in the surgery area allow to conclude that the selected surgical technique was safe and effective. Postoperative radiographic controls are critical, as well as an interdisciplinary team collaboration to carry on the indicated treatment plan.

Conclusions

The incidence of odontogenic tumors of the dental follicle is higher during childhood, and most cases develop without clinical signs or symptoms. An early clinical and radiographic diagnosis, together with an interdisciplinary management, are critical for a timely referral and early treatment of patients who are still growing, thus avoiding late consequences.

In this clinical case, the radical treatment consisted in complete lesion excision and extraction of the unerupted tooth and these corroborated the absence of lesion recurrence or malignant transformation, as evidenced in clinical and remote X-ray controls.

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