

Finding an Ameloblastic Fibro-Odontoma in a Pediatric Patient who Attended a Consultation at the HIGA Pte Peron

Jonathan Bavaro¹, Sebastian Miguelez², Ariel Monteagudo², Marta Peluso³ and Christian Oscar Mosca^{4*}

¹Resident R3 of the Interzonal General Hospital of Agudos Pte. Perón, Buenos Aires, Argentina

²Dentist of the Interzonal General Hospital of Agudos Pte. Perón, Buenos Aires, Argentina

³Head of the Dentistry Service Interzonal General Hospital of Agudos Pte. Perón, Buenos Aires, Argentina

⁴Teacher Collaborator Advisor Interzonal General Hospital of Agudos Pte. Perón, Professor of the Specialty of Surgery and Traumatology Bucomaxillofacial, Faculty of Dentistry of the National University of the Northeast, Corrientes, Argentina

***Corresponding Author:** Christian O Mosca, Teacher Collaborator Advisor Interzonal General Hospital of Agudos Pte. Perón, Professor of the Specialty of Surgery and Traumatology Bucomaxillofacial, Faculty of Dentistry of the National University of the Northeast, Corrientes, Argentina.

Received: February 12, 2019; **Published:** February 28, 2019

Abstract

Odontogenic neoplasms or odontogenic tumors are unique in the oral cavity and do not occur anywhere else in the body. The ameloblastic fibro-odontoma (AFO) is a benign odontogenic mixed tumor of rare appearance.

In this article, we will present the clinical situation of a male patient of 12 years of age who attends the Dental Service of H.I.G.A. Pte. Perón presenting an ameloblastic fibro-odontoma (AFO) in the retromolar trigone area, its surgical removal and its remote control.

Keywords: Ameloblastic Fibro-odontoma; Benign Tumor; Pediatrics; Tumors of the Jaws

Introduction

Odontogenic neoplasms or odontogenic tumors are unique in the oral cavity and do not occur anywhere else in the body. They originate from tissues involved in odontogenesis, that is, derive from the epithelial and mesenchymal remnants of the dental germ, and include a wide variety of lesions ranging from hamartomas to non-neoplastic tissue proliferations and both benign and malignant neoplasms [1].

A tumor is an abnormal tissue mass, with an almost autonomous growth that exceeds that of normal tissues; these formations can be classified into two main categories, benign tumors and malignant tumors.

They are aggressive entities locally and their treatments are quite different from each other. Its frequency of presentation varies according to geographical region, ethnic group, gender, among others [2].

Many clinical studies indicate that odontogenic tumors correspond to 1.8% of the total of oral and tumor lesions that affect the jaws and that the average age of presentation of benign tumors is 25.2 years, specifically 24.5 years for males and men and 25.8 years for females [3].

Clinically, most are located intraosseously, with non-specific symptomatology of slow growth, painless and expansive. Its radiographic appearance is that of radiolucent, radiomix or radiopaque images. Immunohistochemical and cytogenetic advances have helped to further understand their behavior and, in some cases, to include entities that were previously not considered.

It is from these tissue remnants that the formation of tumor masses in the jaws begins.

In this article, we will present the clinical situation of a male patient of 12 years of age who attends the Dental Service of H.I.G.A. Pte. Perón presenting an Ameloblastic Fibro-odontoma (AFO) in the retromolar trigone area, which according to the recent classification of 2017 of the WHO, is grouped in the type of benign mixed odontogenic epithelial and mesenchymal tumors.

The ameloblastic fibro-odontoma (AFO) is a benign mixed odontogenic tumor (epithelial tissue containing ectomesenchyme, with the formation of hard dental tissue) of rare appearance, which constitutes 2% of all odontogenic tumors. The WHO has defined it as a lesion similar to the ameloblastic fibroma (AF) that shows inductive alterations which lead to the formation of enamel and dentin [4,5]. Initially the AFO was denominated, like "ameloblastic odontoma", but, due to its exceptional mixed nature of growth and the incidence of two types of odontogenic tumors with diverse histology and biological behavior, the World Health Organization (WHO) suggested to this term as inappropriate. Later, it is aptly named as an ameloblastic fibro-odontoma (AFO) by Hooker (1967) [6]. It tends to appear more frequently in the jaw (80% to 90%) and in later sectors of young patients (with an average age between 14 and 15 years) without a predilection for sex, sometimes associated with an included tooth. The AF and AFO are considered the same process because they are variants of the same tumor, only differentiated by the presence of an odontoma in the case of AFO. Radiologically, AF is a completely radiolucent and well-defined lesion, whereas AFO is radiolucent with different degrees of radiopacity due to the presence of odontomas. For some authors, the lesion sometimes mimics a complex odontoma appearance [7].

Clinically, the patient may have delayed tooth eruption accompanied by painless volume increase, asymmetric face, there may be some displaced or impacted tooth piece. The mucosa that covers the lesion is normal, has a low tendency to infection and is usually associated with unerupted teeth [8,9].

The treatment varies according to its size and special care must be taken in the decision to keep adjacent teeth or not, since there is a possibility of recurrence of the injury.

The treatment of choice for this lesion consists of enucleation through conservative surgery, followed by curettage, and with post-operative control prolonged over time. The prognosis is excellent, presenting a low recurrence rate [10].

Materials and Methods

For the present investigation fundamentally the rights of the patient were protected, firstly under the consent signed by the mother of the represented and the authorization in the teaching area of the Interzonal General Hospital of Agudos, President Perón, respecting the ethical principles based on the Declaration of Helsinki.

Results

Clinical situation

Interconsultation of 12-year-old pediatric patient with orthodontic reason is received to locate tooth piece 23 retained in the arch. The patient attended with a panoramic x-ray 20 months ago and with a current panoramic radiograph. In the old panoramic radiograph (Figure 1), nothing relevant is observed, but in the current image, by radiographic finding (Figure 2), a radiolucent circumscribed lesion associated with hyperosseous intraosseous zone is observed in the region of the right retromolar triangle; likewise, denticle is also observed between apices of teeth 22 and 24.

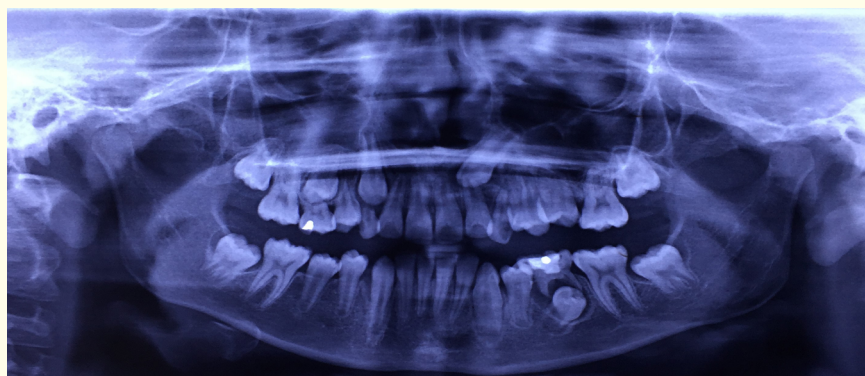


Figure 1: Ancient Panoramic Radiographic Examination 20 months ago. Where the part 23 retained is observed. No pathological entity is observed at the height of the right retromolar trigone.



Figure 2: Current Panoramic Radiographic Examination where the retention of the tooth 23 is observed with the formation of a denticle (supernumerary) between the pieces 22 and 24. In turn a radiolucent image accompanied by a radiopaque image at the height of the trigone retromolar (ameloblastic fibro-odontoma).

Clinically mucosa of normal coloration is observed, the bone tables are kept preserved, the patient does not report painful symptoms. There is delay in the eruption of piece 23, and piece 47 in the process of eruption (Figure 3).



Figure 3: Intraoperative oral examination. Semi-retention of piece 47 can be observed. There are no changes of mucosal coloration or symptoms in the region of the retromolar trigone where the AFO is located.

Complementary studies

Routine blood cell studies, leukocyte formula and basic coagulogram are indicated.

Axial computed tomography of dental piece region 48 is indicated with axial cuts every 1.2 mm, where a circumstantial image of 9 mm diameter (approximately) is observed in area of tooth 48, together with a radiopaque image (Figure 4).

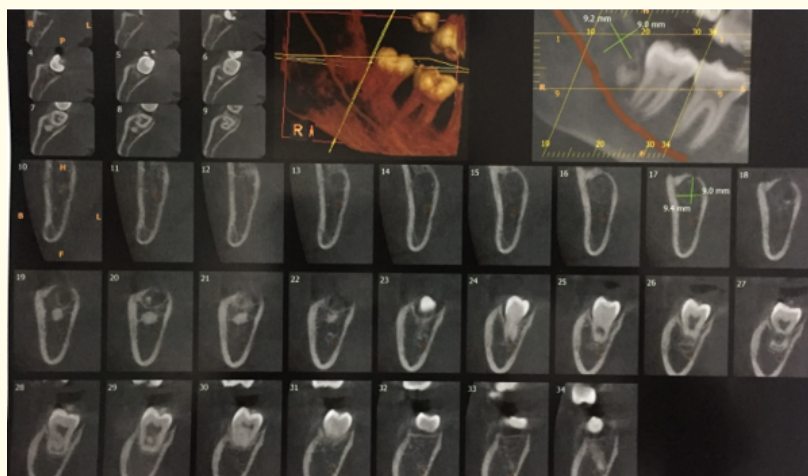


Figure 4: Computed Axial Tomography Cone beam where a circumstantial image of 9 mm diameter (approximately) is observed in area of tooth 48 (retromolar trigone), together with a radiopaque image.

Surgical technique

In order to reduce the amount and activity of the oral microbiota, friction antiseptics were performed in the area of the surgical approach with 10% povidone iodine solution. Subsequently, a truncal anesthesia was performed to the inferior and lingual alveolar nerve, together with infiltrative anesthesia in the vestibular groove bottom for the buccal nerve. Relevant surgical maneuvers were performed to access the pathological entity, making an incision of Newman's medium from the anterior edge of the ascending branch and mesial discharge of piece 46 with cold scalpel blade No. 15c; performing a lingual flap traction point, to facilitate visualization of the area to be operated (Figure 5). The relevant osteotomy is performed with a gouge and strawberry for bone, and the incisional biopsy of the cystic lesion is performed using a Halsted clamp (Figure 6 and 7). The resected piece was immersed in 10% formaldehyde and sent to the Chair of Pathological Anatomy of the Faculty of Dentistry of the University of Buenos Aires.

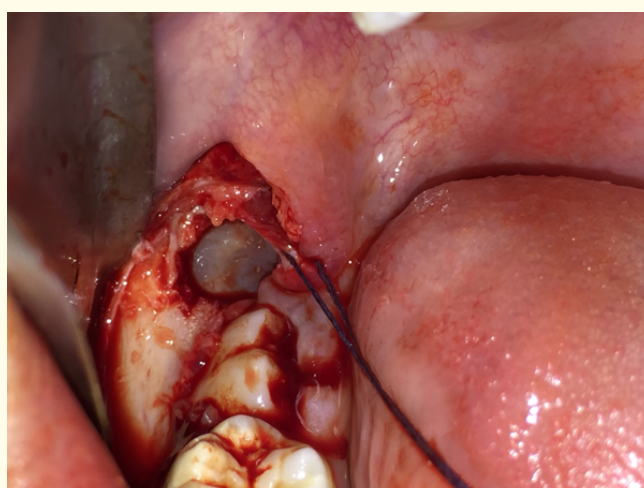


Figure 5: Approach by incision on edge of mandibular ascending branch, intracrevicular with mesial discharge in piece 46. Osteotomy. The whitish grayish color of the pathological entity can be visualized.



Figure 6: Taking a sample for biopsy with Halsted clamp.

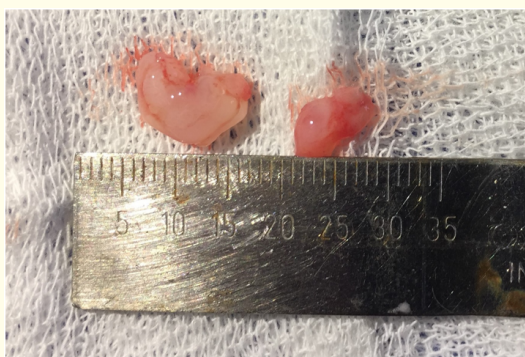


Figure 7: Fragments obtained from the lesion for delivery to the department of pathological anatomy.

Pathological studies

Two fragments of soft tissue measuring 1.2 x 0.7 x 0.1 cm and 0.9 x 0.8 x 0.2 cm, of irregular and whitish surfaces, showing firm consistency to the cut are observed.

The histopathological picture together with the images evaluated correspond to a mixed odontogenic epithelial mesenchymal lesion with formation of dental tissues, resulting in an Ameloblastic fibro-odontoma (Figure 8).

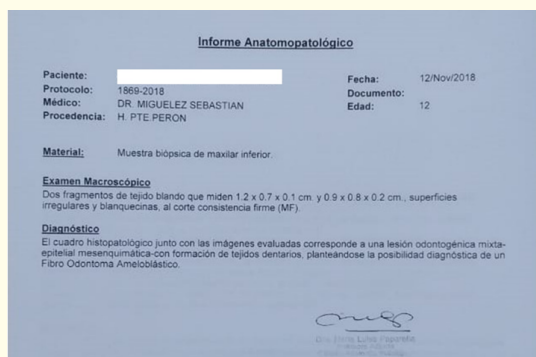


Figure 8: Pathological Anatomy Report resulting in ameloblastic fibro-odontoma (AFO).

Surgical technique. Second surgery

When obtaining an anatomopathological report of the lesion, it was decided to perform the complete exeresis of the cystic entity; for which relevant maneuvers were repeated to access the lesion intraosseously, and be able to remove it completely conservatively, avoiding injury to the Lower Dentaurum (Figure 9 and 10).



Figure 9: Approach to the second surgery. Conservative removal of the lesion.



Figure 10: Remains of the membrane and odontomas. Remission of the second sample confirms the diagnosis of ameloblastic fibro-odontoma (AFO).

A sample of hard tissue of blackish appearance compatible with odontoma and soft tissue in 10% form is sent to be analyzed by the Chair of Pathological Anatomy of the Faculty of Dentistry of the University of Buenos Aires, obtaining the same result as the first biopsy.

Post-surgical controls

After one week, postoperative controls, suture removal and radiographic control are performed, where a small area of greater bone density distal to part 47 is observed. The patient does not manifest having paresthesia of the inferior nerve.

Discussion

Benign odontogenic tumors represent one of the most complex sections among the pathological processes that settle in the oral cavity. Its growth derived from alterations in embryological development will give rise to different forms of neoplasms with different dental tissues.

The origin of these odontogenic tumors occurs because odontogenesis begins during the sixth week of intrauterine life, with the formation of the first teeth from the oral epithelium, which covers the maxillary and mandibular alveolar processes through the intussusception of the ectomesenchyme. It fixes the location of the dental pieces, besides stimulating the growth of the dental lamina as well as the enamel and induce the ameloblasts to the formation of enamel by means of the differentiation of the odontoblasts and finally the one of arriving to differentiate the mesenchymal cells in cementoblasts. During this process of dental training tend to be remnants of the following structures:

- Remains of the dental lamina or remnants of Serres.
- Remains of the enamel organ.
- Remains of the Hertwing sheath or remains of Malassez.

The primary tumors of the maxillofacial region in pediatric patients are infrequent in comparison to those that occur in the adult, and the damage that these injuries cause in the tissues, is of greater impact, since they directly alter the growth and facial development, as the psychosocial development of children. In general, the tumor lesions in children usually show an aggressive local behavior, being complicated the initial diagnosis and plan of later treatment, having to determine the degree of malignancy and the histological lineage of the tumors, since the treatment must go directed to the resection of the lesion and the immediate reconstruction of the tissues, if possible, restoring function and aesthetics in a single procedure, as well as favoring the growth of the affected structures. All this is possible when the lesions are of benign origin, and can hardly be performed when they are of malignant origin. Whichever the case, it is difficult to reach an initial diagnosis, since signs and symptoms, as well as radiographic findings are sometimes similar or nonspecific for each lesion, being able to confuse degree of malignancy or histological origin, being for this, the taking of biopsy the most important element for a correct diagnosis [11].

The AF and AFO are considered the same process because they are variants of the same tumor, only differentiated by the presence of an odontoma in the case of AFO. Radiologically, AF is a completely radiolucent and well-defined lesion, whereas AFO is radiolucent with different degrees of radiopacity.

The differential diagnosis of AF should be made with lesions such as ameloblastoma, odontogenic myxoma, dentigerous cysts, odontogenic keratocysts, central giant cell granuloma and histiocytosis. In addition, the AFO should be distinguished from calcifying epithelial odontogenic tumor (Pindborg tumor) and calcifying odontogenic cyst (Gorlin cyst), odontomas, adenomatoid odontogenic tumor (AOT), juvenile ossifying fibroma and ossifying cement fibroids. Most recurrences are attributed to incomplete resections; thus, Zallen, *et al.* reviewed the medical literature in 1982 and found 85 cases of AF, of which 14 (18.3%) had recurred [12]. In addition, cases of malignant transformation of AF and AFO have been described in ameloblastic fibrosarcoma (44% of the fibrosarcomas described). Therefore, there is controversy regarding the form of treatment, although most authors recommend an adequate resection followed by a vigorous surgical bed curettage [13-16].

The origin of this pathology at the molecular level is attributed to the alteration in the b-catenin and CD44 pathway that has been shown to be actively involved in the development of other odontogenic tumors, such as ameloblastoma [17].

Conclusion

Tumors of primary origin in children often show rapid local growth due to the growth potential of the cells, with local invasion and tissue destruction, which in many cases do not correlate with their benign histological appearance. Despite this rapid and destructive behavior, injuries in children are usually benign, but treatment must be based on their clinical and biological behavior.

It is important to take into account diagnostic aids such as laboratory tests, x-rays, computerized or three-dimensional tomography, magnetic resonance or even ultrasound, being elementary to guide the diagnosis, although these studies are inconclusive.

In our situation, based on the literature that indicates the low recurrence of this entity, the treatment was conservative.

Bibliography

1. Jing W., *et al.* "Odontogenic tumours: a retrospective study of 1642 cases in a Chinese population". *Journal of Oral and Maxillofacial Surgery* 36.1 (2008): 20-25.
2. Fernandes A., *et al.* "Odontogenic tumors: a study of 340 cases in a Brazilian population". *Journal of Oral and Maxillofacial Surgery* 34.10 (2005): 583-587.
3. Da-Costa DO., *et al.* "Odontogenic tumors: a retrospective study of four Brazilian diagnostic pathology centers". *Medicina Oral Patologia Oral y Cirugia Bucal* 17.3 (2012): 389-394.
4. Kruse A. "Über Die Entwicklung Cystischen Gesschwulse in Unterkiefer". *Archiv für Pathologische Anatomie und Physiologie* 124 (1891): 137.
5. Mosby EL., *et al.* "Ameloblastic fibroma in a 7-week-old infant: A case report and review of the literature". *Journal of Oral and Maxillofacial Surgery* 56.3 (1998): 368-372.
6. Hooker SP. "Ameloblastic odontoma: An analysis of twenty six cases". *Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology* 24.3 (1967): 375.
7. Ghandehari-Motlagh M., *et al.* "Fibro-odontoma amelobástico en un niño de 4 años". *Revista Iraní de Pediatría* 26.2 (2016): e3124.
8. Anshad Mohamed Abdulla., *et al.* "Ameloblastic Fibroodontoma: Uncommon Case Presentation in a 6-Year-Old Child with Review of the Literature". *Case Reports in Medicine* (2017): 9483738.
9. Singh AK., *et al.* "Fibroodontoma u odontoma ameloblastico: dos caras de la misma moneda". *National Journal of Maxillofacial Surgery* 7.1 (2016): 92-95.
10. Castellón M L., *et al.* "Fibro-odontoma ameloblástico de la mandíbula". *Revista Española de Cirugía Oral y Maxilofacial* 35.2 (2013): 87-92.
11. Montañez FM. "Aggressive mandibular tumors in pediatric patients. Report of 4 cases". *Revista Odontológica Mexicana* 20.2 (2016): e125-e131.
12. Zallen RD., *et al.* "Ameloblastic fibroma". *Journal of Oral and Maxillofacial Surgery* 40.8 (1982): 513-517.
13. Martín-Granizo-López R., *et al.* "Fibroma ameloblástico mandibular. Presentación de dos casos". *Medicina Oral Patologia Oral y Cirugia Bucal* 8 (2003): 150-153.
14. Contreras William., *et al.* "Peripheral Developing Odontoma or Peripheral Ameloblastic Fibro-Odontoma Erupting to Oral Cavity Case Report". *International Journal of Odontostomatology* 12.2 (2018): 117-120.
15. Singh AK., *et al.* "Ameloblastic fibroodontoma or complex odontoma: Two faces of the same coin". *National Journal of Maxillofacial Surgery* 7.1 (2016): 92-95.
16. Mikami T., *et al.* "Congenital peripheral developing odontoma accompanied by congenital teratomatous fibroma in a 9-month-old boy: a case report". *Journal of Oral Science* 55.1 (2013): 89-91.
17. Lin Y., *et al.* "Ameloblastoma with varied sites of metastasis: Report of two cases and literature review". *Journal of Cranio-Maxillofacial Surgery* 42.5 (2014): e301-e304.

Volume 8 Issue 3 March 2019

©All rights reserved by Christian Oscar Mosca., *et al.*