Recurrence of Ameloblastic Fibro-Odontoma in a Child: A Case Report

Raphael Capelli Guerra, DDS, MSc1,2, Pedro Henrique de Azambuja Carvalho, DDS1,2, Florian Markus Thieringer, DDS, MD, MSc3,4, Rodrigo Santos Pereira, DDS, MSc, PhD5, and Eduardo Hochuli-Vieira, DDS, MSc, PHD1,2

Abstract
Introduction: A young boy presented with mandibular swelling at the Department of Oral and Maxillofacial Surgery in Hospital Leforte, São Paulo, Brazil. Radiography showed a mixed bone lesion. Computed tomography (CT) findings were compatible with ameloblastic fibro-odontoma (AFO).

Case Report: An excisional biopsy was performed under general anesthesia. At the 6-year follow-up, local inflammation and mild exudates were found. Local recurrence of the lesion was observed on CT. Therefore, excision and curettage were performed. No signs of recurrence have been detected to date. Ameloblastic fibro-odontoma is a rare benign neoplasm most commonly occurring in the first decade of life, with minimal probability of malignancy.

Conclusion: Clinical, radiological, and histopathological findings aid in the diagnosis, and long-term follow-up is necessary as recurrence can develop even after 6 postoperative years.

Keywords
ameloblastic fibro-odontoma, impacted tooth, recurrence, case report

Received: 27 May 2019; accepted: 7 January 2020

Introduction
Ameloblastic fibro-odontoma (AFO) is a relatively rare mixed radiopaque/radiolucent tumor. It accounts for 0.3% to 1.7% of the cases of odontogenic tumors.1 AFO lesions have unique features and are rarely recurrent.1,2

Ameloblastic fibro-odontoma is a rare benign mixed odontogenic tumor composed of soft and hard tissues.3 The soft tissue is predominantly the odontogenic epithelium and odontogenic mesenchyme, while the hard tissue has foci of dentin and enamel.3,4 It occurs most commonly in the first two decades of life.2

Ameloblastic fibro-odontoma is treated with enucleation, and the prognosis is excellent in children with small and asymptomatic lesions. For severe cases, radical treatments may be performed, but without involving the adjacent teeth.5-7

1 Department of Diagnostic and Maxillofacial Surgery, School of Dentistry (UNESP) Araraquara, São Paulo State University, Araraquara, São Paulo, Brazil
2 Department of Oral and Maxillofacial Surgery, Hospital Leforte, São Paulo, Brazil
3 Department of Oral and Maxillofacial Surgery, University Hospital Basel, Basel, Switzerland
4 3D Printing Lab Department, University of Basel, Basel, Switzerland
5 Department of Oral and Maxillofacial Surgery, São Paulo State University, School of Dentistry (UNESP) Araçatuba, Araçatuba, Brazil

Corresponding Author:
Raphael Capelli Guerra, DDS, MSc, Department of Diagnostic and Maxillofacial Surgery, School of Dentistry (UNESP) Araraquara, São Paulo State University, Araraquara, São Paulo 14801903, Brazil.
Email: dr.raphael.guerra@gmail.com

Creative Commons Non Commercial CC BY-NC: This article is distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 License (https://creativecommons.org/licenses/by-nc/4.0/) which permits non-commercial use, reproduction and distribution of the work without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (https://us.sagepub.com/en-us/nam/open-access-at-sage).
Here, we report a rare case of recurrent AFO in a young child at the 6-year follow-up.

**Case Report**

In 2011, a 6-year-old boy presented with a slight intraoral swelling of the external cortex of the mandible to the Department of Oral and Maxillofacial Surgery in University Hospital Leforte, São Paulo, Brazil. Radiography showed a mixed bone lesion, with involvement of the germ of the mandibular right third molar and mandibular right second molar. Computed tomography (CT) was performed to analyze the affected areas and evaluate the depth of the lesion. A histopathological analysis of an incisional biopsy specimen showed a mixed lesion compatible with AFO.

The entire lesion, including the tooth germs, was removed under general anesthesia. The removal resulted in a bone defect at the angle of the mandible on the right side (Figure 1A). Due to brittleness in the region of the bone defect, the defect was filled with bone marrow obtained from the iliac crest. Fixation was performed with absorbable inion plates.

There were no postoperative complications. On postoperative day 37, the patient presented with local dehiscence in a small area in the suture region, which was treated with saline irrigation.

No abnormalities were detected for 6 years of follow-up. However, at the 6-year follow-up, he presented with local unevenness of bone thickness, exposed bone tissue, and mild local inflammatory exudates in the region of the retromolar space on the right side. A new lesion was detected on CT (Figure 2).

Curettage was performed under general anesthesia to remove the lesion and adjacent soft tissues. A histopathological examination confirmed the relapse of AFO. No abnormalities were detected at the 6-month follow-up.

**Discussion**

The benign odontogenic tumor AFO is classified by the World Health Organization (WHO) as a rare tumor that exclusively affects children at a mean age of 6.3 years, as in the present case. AFO is not aggressive, so it is not necessary to remove the adjacent oral tissues. Clinical, radiographic, and histopathologic features aid in the diagnosis.

Recurrence of AFO is also rare and occurs in approximately 6% of the cases. The age of the present patient was compatible with that reported in the literature for the development of AFO, and relapse occurred after 6 years. A systematic review showed that frequencies of AFO in the mandibular and maxillary regions were 65% and 35%, respectively.

Howell & Burkes (1977) reported two cases of relapse of AFO and cellular differentiation of AFO to fibro-odontosarcoma. However, such differentiation to malignancy is rare, and the treatment for these cases is chemotherapy and radiotherapy combined with extensive surgical resection.

Ameloblastic fibro-odontoma is the histological combination of ameloblastic fibroma (AF) and complex odontoma. It shows the same benign histological behavior as AF. In contrast, ameloblastic odontoma is the histological combination of complex ameloblastoma and odontoma, which has the same invasive behavior as ameloblastoma.

There are controversies on the classification of AFO into AF and ameloblastic fibro-dentinoma. In 1971, the WHO defined AF as a benign neoplasm composed of proliferation of the odontogenic epithelium, interspersed with ectomesenchyme, which resembles the ectomesenchyme of the dental papilla. AF involving the dentin and enamel progresses to AFO. In the 2005 classification, AFO, AF, and ameloblastic fibro-dentinoma were defined as separate entities, and the possibility of malignant transformation of AF to ameloblastic fibrosarcoma was highlighted. Finally, in the 2017 classification, an association among the three pathologies was reported: AF and ameloblastic fibro-dentinoma were defined as the progression of AFO. Furthermore, it was reported that the malignant transformation of AF is rare.

Consensus among the authors on the first-choice treatment for neoplasia is total enucleation. In cases of relapse...
or malignant potential, resection with a wide margin or marginal mandibulectomy is recommended. Long-term radiographic monitoring is recommended because of the possibility of recurrences.

Conclusion
A long-term follow-up of AFO is necessary because of the possibility of recurrence, and an early diagnosis should be provided to avoid radical procedures.

Declaration of Conflicting Interests
The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding
The author(s) received no financial support for the research, authorship, and/or publication of this article.

ORCID iDs
Raphael Capelli Guerra, DDS, MSc https://orcid.org/0000-0002-9071-7827
Pedro Henrique de Azambuja Carvalho, DDS https://orcid.org/0000-0003-2670-7998
Florian Markus Thieringer, DDS, MD, MSc https://orcid.org/0000-0003-3035-9308

References
1. Sardana D, Rattan V, Gauba K, Goyal A, Singh S, Vai-Phei K. Surgical excision of large ameloblastic fibro-odontoma followed by alloplastic bone graft—2 year follow-up: a case report. J Oral Maxillofac Surg Med Pathol [Internet]. 2015;27(1):41-44.
2. Kirjavainen A, Tuovinen V, Sándor G. Large ameloblastic fibro-odontoma in a 7-year-old girl with analysis of 108 cases. Ann Maxillofac Surg [Internet]. 2016;6(1):15.
3. Reichart PA, Philipsen HP, Gelderblom HR, Stratmann U. Ameloblastic fibro-odontoma—report of two cases with ultrastructural study of tumour dental hard structures. Oral Oncol Extra. 2004;40(1):8-12.
4. Buchner A, Kaffe I, Vered M. Clinical and radiological profile of ameloblastic fibro-odontoma: an update on an uncommon odontogenic tumor based on a critical analysis of 114 cases. Head Neck Pathol. 2013;7(1):54-63.
5. Ülgür II, Caduff R, Erb J, Van Waes H, Jacobsen C, Bredell MG. Ameloblastic fibro-odontoma located in the maxilla of a 3-year-old girl. *Pediatr Dent J*. 2014;24(2):106-110.

6. Mainenti P, Oliveira GS, Valério JB, et al. Ameloblastic fibro-odontosarcoma: a case report. *Int J Oral Maxillofac Surg*. 2009;38(3):289-292.

7. Surej Kumar LK, Manuel S, Kalam SA, Venugopal K, Sivakumar TT, Issac J. Ameloblastic fibro-odontoma. *Int J Surg Case Rep* [Internet]. 2014;5(12):1142-1144.

8. Howell RM, Burkes EJ. Malignant transformation of ameloblastic fibro-odontoma to ameloblastic fibrosarcoma. *Oral Surg Oral Med Oral Pathol*. 1977;43(3):391-401.

9. Gilani SM, Raza A, Al-Khafaji BM. Ameloblastic fibrosarcoma: a rare malignant odontogenic tumor. *Eur Ann Otorhinolaryngol Head Neck Dis* [Internet]. 2014;131(1):53-56.

10. De Riu G, Meloni SM, Contini M, Tullio A. Ameloblastic fibro-odontoma. Case report and review of the literature. *J Cranio-Maxillofacial Surg* [Internet]. 2010;38(2):141-144.