

Maxillary Ameloblastic Fibroma: A Case Report

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Ameloblastic fibroma is a relatively rare benign odontogenic tumor in which both the epithelial and ectomesenchymal components are neoplastic. An 8-year-old Caucasian boy was referred to the dentist for evaluation of failed eruption of the maxillary left first molar. The panoramic radiograph showed a well-circumscribed unilocular radiolucency involving an unerupted maxillary left first permanent molar. The lesion was enucleated and the material was sent for histopathologic examination. Microscopically, it was composed by cords and islands of odontogenic epithelium in a myxoid cell-rich stroma that closely resemble the dental papilla with histopathological diagnosis of ameloblastic fibroma. After 24 months of follow-up no recurrence was observed and the maxillary left first molar erupted spontaneously through the buccal mucosa and was aligned with a fixed orthodontic appliance. This case emphasized the importance of careful differential diagnosis of intraosseous oral lesions and reported a rarity of the lesion and its atypical location.

Key Words: ameloblastic fibroma, odontogenic tumor, maxillary tumor.

INTRODUCTION

Ameloblastic fibroma (AF) is a benign odontogenic tumor considered a true mixed tumor composed of neoplastic epithelium and mesenchymal (1). This tumor is rare, representing only 2% of all odontogenic tumors (1-5).

AF has been reported in patients aged 7 weeks to 51 years (6,7), but the tumor is considered a tumor of childhood and adolescence and occur almost exclusively in the first and second decades of life (6,8-13). In the majority of cases, the lesion arises in the mandible and patients present as a slow-growing, painless lesion and/or failure of the tooth eruption (8-12). However, the tooth impaction and delayed eruption may be associated with dental developmental defects or abnormalities such as amelogenesis imperfecta (14). In other cases, the tumors are asymptomatic and discovered during routine oral/radiographic examination (12).

Radiographically, AF appears as well-defined unilocular or multilocular radiolucencies (2,7,10). It has

been reported that asymptomatic cases present unilocular radiolucency, whereas multilocular cases are associated with jaw swelling (15). However, if the lesion contains radiopaque masses in the internal structure, the diagnosis may be calcifying epithelial odontogenic tumor (16).

There are several variants of AF. If dentin is present, the tumor is ameloblastic fibrodentinoma. If dentin and enamel are present, the tumor is called ameloblastic fibro-odontoma (AFO) (15,17). Mitotic figures may be present in AF. The presence of a large number of cells in mitosis and atypical mitosis should expand the differential diagnosis to include malignant entities, like ameloblastic fibrosarcoma (1).

The most appropriate treatment method for AF is still uncertain. A conservative approach is suggested by several authors (1,10,11). However, tumors may recur following surgical removal and progress to malignancy (7,18,19). Thus, long-term follow-up of AF is recommended (18). This report describes an interesting case of AF that affected the maxilla of a young boy and was associated with an unerupted maxillary left first molar.

CASE REPORT

In May 2008, an 8 year-old Caucasian boy was referred to the dentist for evaluation of failed eruption of the permanent maxillary left first molar. Physical examination showed a generally healthy child and the medical, surgical, family, and social histories were unremarkable.

Intraoral examination revealed a small painless expansion of the buccal cortical maxillary plate and the first molar was not visualized. The overlying mucosa was intact and normal in color and consistency.

Radiographic examination by means of a panoramic radiograph (Fig. 1A) showed a well-defined unilocular radiolucent lesion in the left maxilla. The crown of the permanent first molar was apparent within the lesion. The second molar was displaced upwards. Computed tomography (CT) scanning confirmed the findings of the panoramic radiograph (Fig. 1B). Based on clinical and roentgenographic findings, a presumptive preoperative diagnosis of dentigerous cyst was done.

Under local anesthesia, the lesion was totally removed through an intraoral approach and protecting the definitive first molar. The enucleated material was sent to the Anatomic Pathology Service of the Antonio Pedro University Hospital, Niterói, RJ, Brazil for histological examination.

Microscopically, the lesion consisted of cords and islands of proliferating odontogenic epithelium. The cords often revealed a double or triple layer of cuboidal cells, resembling the dental lamina. The

island showed peripheral rim of columnar similar to the inner enamel epithelium. The center showed a loosely arranged spindle-shaped epithelium identical to stellate reticulum. The epithelial components laid in a myxoid cell-rich stroma with little collagens closely resembling the primitive dental papilla. No hard tooth structures or mitotic images were noted. The histopathological diagnosis was AF (Fig. 2).

After 24 months of follow-up, no recurrence was observed. The first molar was subjected to traction and was aligned with a fixed orthodontic appliance (Fig. 3A). However, radiographic examination revealed that the second molar was impacted to the first molar (Fig. 3B).

DISCUSSION

AF is a mixed tumor both epithelial and mesenchymal neoplastic proliferation (1). This tumor is usually diagnosed between the first and second decades of life and the majority of cases occur in the mandible (2,10,12,15). However, few cases of the AF in the maxilla have been reported, as the case presented in this paper (9,10,13).

The clinical manifestations of AF are not specific and the lesion is frequently discovered as an incident finding in a routine radiographic examination, such as cysts and other odontogenic tumors (1,9). Radiographically, the differential diagnosis of the AF must be made with entities such as dentigerous cysts, ameloblastoma, odontogenic keratocysts (9) and fibrosarcoma ameloblastic (11,18).

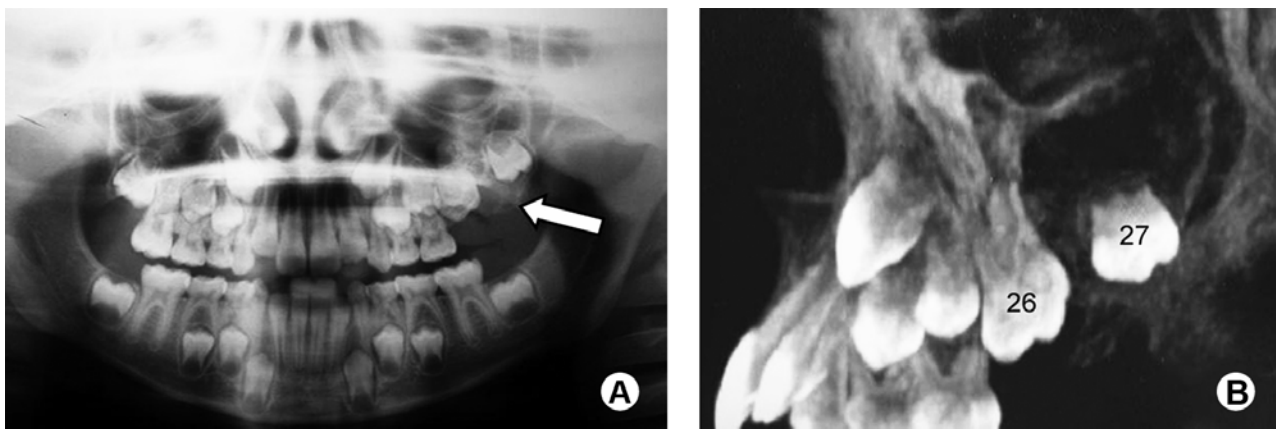


Figure 1. Imaging. A= Initial panoramic film showing a well-circumscribed unilocular radiolucency involving an unerupted permanent maxillary left first molar and displacement of the maxillary left second molar (arrow). B= Computed tomography (CT) scan confirming the findings of the panoramic radiograph.

Philipsen et al. (17) managed AF by enucleation with careful clinical and radiographic follow-up. AF recurrence may not be true recurrence, but rather a residual lesion due to incomplete enucleation. Some authors believe that an aggressive treatment is not justified (2,15,17). However, Chen et al. (7) reported that patients treated by radical excision experience fewer recurrences compared to those treated conservatively. Those authors found recurrence in 14 of 41 cases. Dallera et al. (2), on the other hand, reported no recurrences for 6 cases with an average follow-up of 15 years. In the present case, the lesion was enucleated with no recurrence at two year.

While uncommon, the possibility of malignant

transformation of AF into ameloblastic fibrosarcoma has been reported by Chen et al. (7) and Kobayashi et al. (18). For those authors, AF showed no signs of malignancy until the second recurrence. Chen et al. (7) reported that the rates of malignant transformation in 5-year and 10-year follow-up were 10.2% and 22.2%, respectively. However, according to Kousar et al. (19) rapid sarcomatous transformation of an AF occurred within 6 months.

In the case reported here, the main feature was the failure of eruption of a maxillary first molar. This disturbance may or may not be associated with a pathology, such as AF. Thus, this case emphasizes the importance of careful differential diagnosis in dentistry, while reporting a rare lesion and its atypical location.

RESUMO

Fibroma ameloblástico é um tumor odontogênico benigno relativamente raro, em que ambos os componentes epiteliais e ectomesenquimais são neoplásicos. Menino de oito anos de idade, branco, foi encaminhado ao dentista para avaliar a falha na erupção do primeiro molar maxilar do lado esquerdo. A radiografia panorâmica revelou imagem radiolúcida, unilocular, bem circunscrita, envolvendo o primeiro molar permanente maxilar esquerdo incluso. A lesão foi enucleada e o material encaminhado para avaliação histopatológica. Microscopicamente, era composta de ilhas e cordões de epitélio odontogênico num estroma mixóide rico em células, que se assemelhava à papila dentária, com diagnóstico histopatológico de fibroma ameloblástico. Após 24 meses de acompanhamento, nenhuma recorrência foi observada e o primeiro molar permanente maxilar irrompeu espontaneamente através da mucosa bucal e foi alinhado com aparelho ortodôntico fixo. Esse caso enfatiza a importância do cuidadoso diagnóstico diferencial das lesões orais intra-ósseas e relato de lesão rara e sua localização atípica.

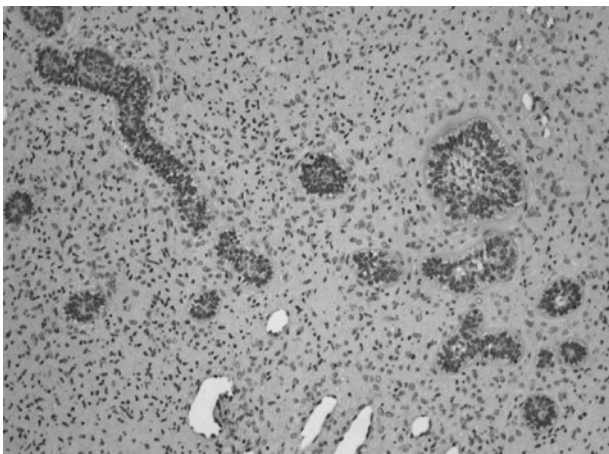


Figure 2. Histological section showing ameloblastic epithelium in cords and island lie in a myxoid cell-rich stroma resembling the primitive dental papilla (HE, ×100 magnification).

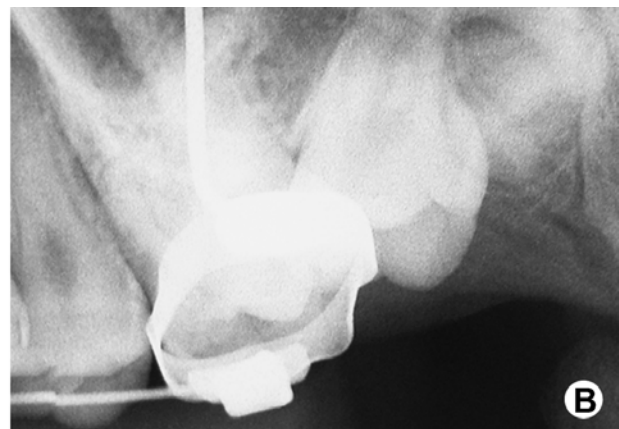
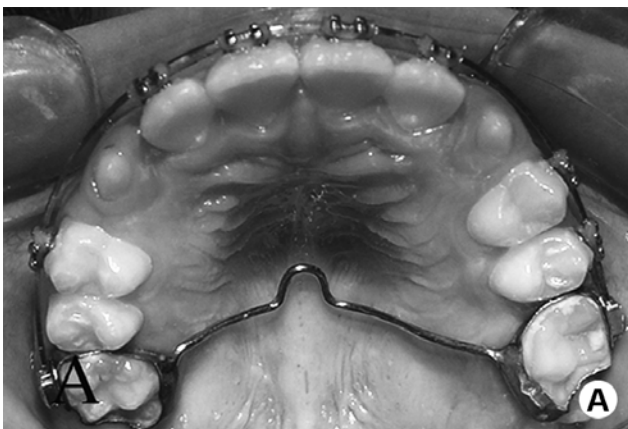


Figure 3. Follow-up. A= Image showing tractioned and aligned the maxillary left first molar with the fixed orthodontic appliance. B= Periapical film showing that the second molar is impacted against the first molar.

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