Diagnosis of ameloblastic fibro-odontoma: case report

Diagnóstico de fibro-odontoma ameloblástico: relato de caso

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Abstract

Objective: the present report describes the clinical, radiographic, and histopathological features of an ameloblastic fibro-odontoma (AFO) lesion. Case report: we report a clinical case of a 14-year-old boy with asymptomatic edema. Panoramic radiography detected a unilocular lesion with defined margins located in the posterior region of the mandible. The internal structure of the lesion presented several degrees of radiopacity with the involvement of the third molar. Cone-beam computed tomography revealed expanded buccal and lingual cortical bones, perforation of the lingual cortical bone, and displacement of the mandibular canal. AFO was suspected based on the radiographic and clinical characteristics. Total excision was performed and histologically examined, confirming the diagnosis of AFO. No recurrence occurred during a 24-month follow-up period. Final considerations: the evaluation of the clinical, radiographic, and histopathologic findings needs to be accurate for a correct diagnosis and appropriate treatment for case of AFO since the presentation is often asymptomatic.

Keywords: odontogenic tumors; radiography panoramic; cone-beam computed tomography.

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Introduction

In 2005, the World Health Organization (WHO) defined ameloblastic fibro-odontoma (AFO) as a benign odontogenic epithelial tumor with ectomesenchyme undergoing variable inductive changes during the formation of the enamel and dentinal matrix[1]. Some ameloblastic fibroma (AF) lesions have neoplastic characteristics and cannot produce the hard dental structures — enamel and dentin — although other AF lesions display morphophysiological changes and start producing hard dental tissues that mature into odontomas. Thus, AFO and ameloblastic fibrodentinoma (AFD) are considered as developing odontomas per the recent WHO classification[2]. However, the classification of AFO has been debated in the literature. In the 2017 WHO classification, this lesion was considered as a developing odontoma; however, few characteristics of this lesion are inconsistent with this progressive maturation[3].

Although AFO is a rare lesion, it occurs more frequently in persons aged below 20 years, with a slightly higher prevalence among men[4]. AFO presents asymptomatically with gradual, expansive growth and occurs more frequently in the posterior region of the mandible. It may lead to delayed eruption, mobility, and tooth displacement[5]. AFO lesions are typically recognized as unilocular masses on radiographic images with defined limits, presenting with variable amounts of calcified material in the internal structure[6]. The histological characteristics of AFO are similar to those of AF, with nests and cords of odontogenic epithelium in an ectomesenchyme as in the dental papilla. Moreover, AFO lesions present with immature mineralized structures, including the enamel and dentinal matrix, which are correlated with odontogenic epithelium[7]. Conservative treatment of AFO comprises surgical removal; in particular, more expansive lesions require wider surgical margins. Treatment has favorable prognosis, and recurrence or transformation into ameloblastic fibrosarcoma is rare[8]. The present study aims to present a clinical case of AFO with an emphasis on its diagnosis.

Case report

A 14-year-old boy presented to the dental clinic of the São Leopoldo Mandic Dental School and Research Center in April 2018 with asymptomatic edema in the left and posterior region of the mandible. The patient denied changes in health and any history of trauma at this site. During the extraoral examination, slight asymmetry was observed on the left region of the face, near the angle of the mandible. This area was hardened, non-fluctuant, and well-circumscribed upon palpation. The intraoral examination showed asymptomatic edema with rigid consistency on palpation, located in the left and posterior region of the mandible, distal to the second molar. The pulp sensitivity test of the second molar revealed no abnormalities; the percussion test revealed no abnormalities and no tooth mobility was observed. The third molar was absent.

An Orthopantomograph® OP300 (Instrumentarium Dental, Tuusula, Finland) device was applied for imaging the lesion. Panoramic radiography revealed a lesion with several degrees of radiopacity in the left mandibular ramus and angle. It was unilocular with defined limits, mostly corticalized, and adjacent to a radiolucent halo. The lesion involved the impacted third molar that was mesio-angled, undergoing inductive changes, and was located near the base of the mandible. The roots of the second molar were displaced to the mesial region with no external root resorptions (Figure 1). On cone-beam computed tomography (CBCT), a hyperdense area delimited above the impacted third molar and a hypodense area involving the third molar and hyperdense area were observed. Displacement of the mandibular canal to the lower cortical region of the mandible was observed (Figure 2). The buccal and lingual cortical bones were expanded, and perforation of the lingual cortical bone was observed, all of which are consistent with the diagnosis of pathological fractures (Figure 3).

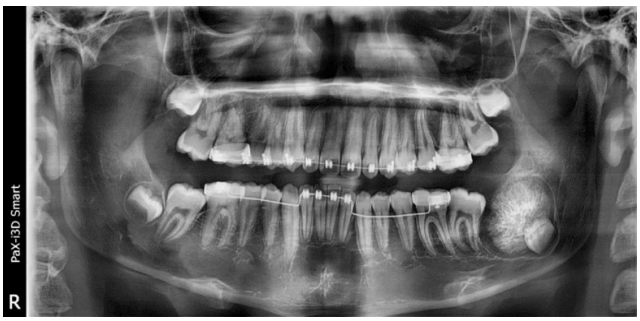

Figure 1 – Panoramic radiography showing a radiopaque image with an adjacent radiolucent halo involving the impacted lower third molar on the left

Source: the authors (SLMandic, Campinas, SP, Brazil).

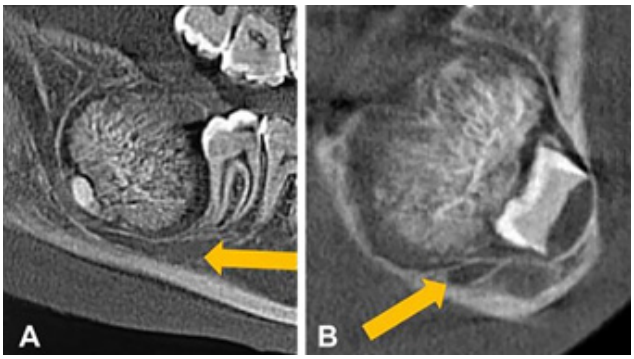

Figure 2 – A. Sagittal and B. Coronal cone-beam computed tomography images showing the inferior displacement of the mandibular canal (yellow arrow)

Source: the authors (SLMandic, Campinas, SP, Brazil).

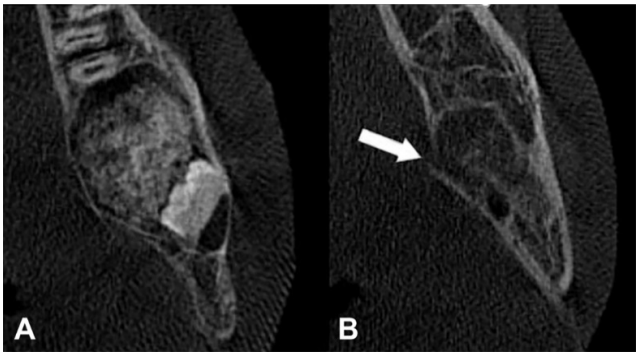

Figure 3 – Axial cone-beam computed tomography images showing A. expanded buccal and lingual cortical bone and B. perforation of the lingual cortical bone (white arrow)

Source: the authors (SLMandic, Campinas, SP, Brazil).

Given the clinical features and radiographic images, the differential diagnosis included lesion such as AFO. Surgery was performed under general anesthesia, with intraoral buccal access on the left mandibular body, with complete lesion removal and the third molar was extracted (Figure 4A). The specimen comprised a tissue mass measuring 38 × 25 × 20 mm with irregular shape and surface, brown coloration, blackened areas, and a hard consistency (Figure 4B). The macroscopic specimen was sent for histopathological analysis, which confirmed the diagnosis of AFO.

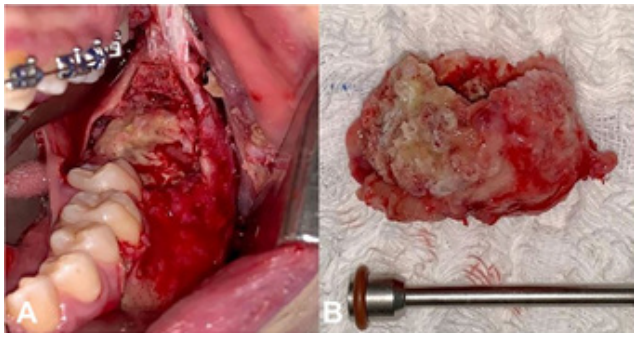

Figure 4 – A. Intraoperative image B. Macroscopic sample

Source: the authors (SLMandic, Campinas, SP, Brazil).

Histologically, the lesion comprised cellularized and loose connective tissue with a basophilic aspect resembling that of the dental papilla. Nests and cords of odontogenic epithelium and structures similar to those of rudimentary dental organs were noted, showing ameloblast-like columnar cells on the periphery, and angled and spaced epithelial cells, similar to the starry reticulum of the enamel, in the innermost sections of these structures (Figure 5A). Most of the lesion was composed of hard dental tissue that was analyzed after demineralization. The analysis revealed a mixture of hard and soft tissue components with sheets of odontogenic epithelial cells permeated by dentin (Figure 5B). In other regions, extensive areas of atubular and tubular dentinal tissues were observed within the nests of odontogenic epithelium (Figure 5C). Well-formed dental structures, consistent with that in teeth formation, exhibiting tubular dentinal and enamel matrix, were also identified in the specimen (Figure 5D).

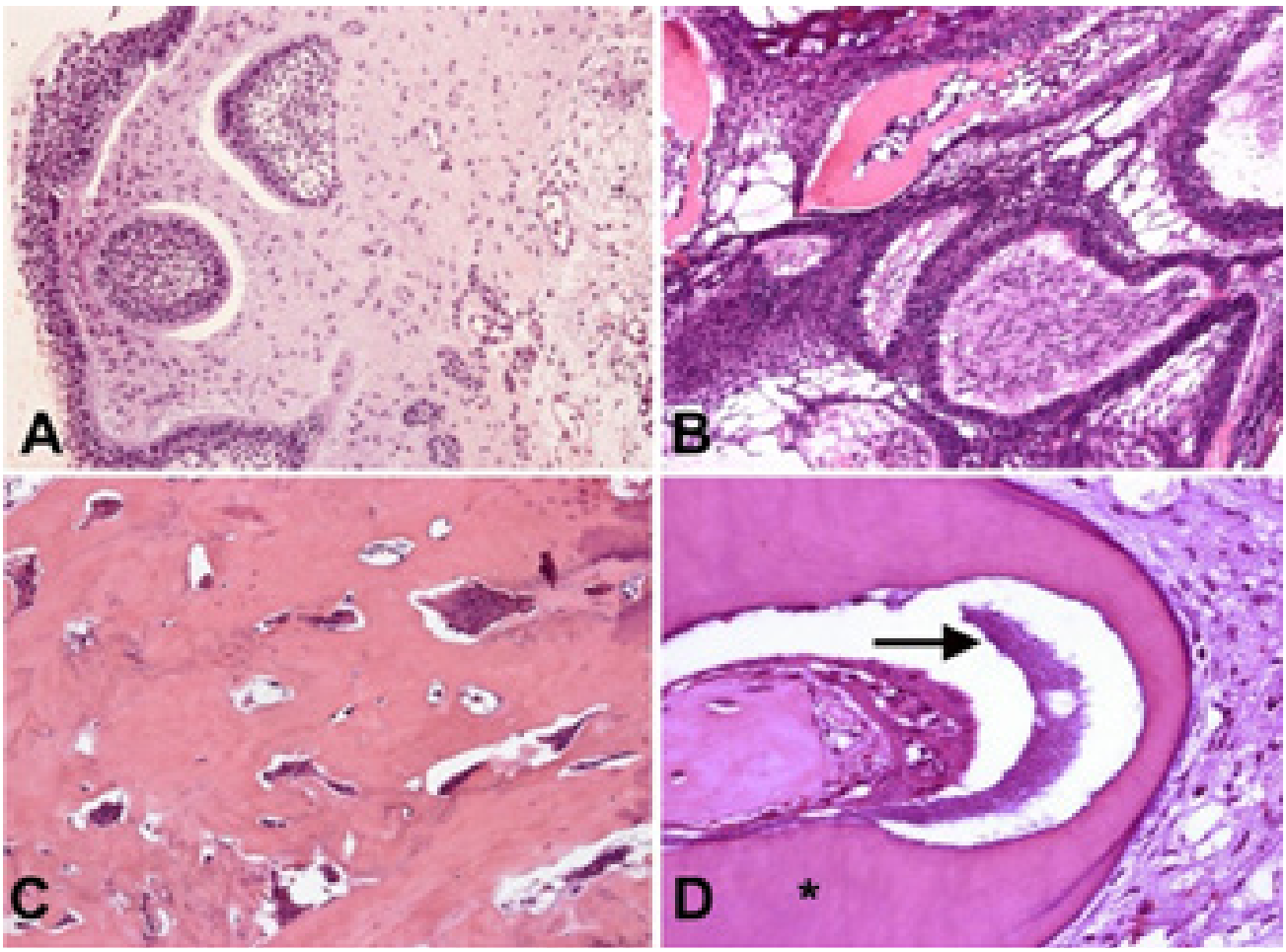

Figure 5 – Histopathological findings: A. The lesion consists of loose, cellularized connective tissue, similar to that of odontogenic ectomesenchyme, with nests and cords of odontogenic epithelium, some of which are organized into structures similar to rudimentary dental germs (hematoxylin and eosin staining, original magnification 200×). B. Sheets of odontogenic epithelial cells permeated by dentin tissue (hematoxylin and eosin staining, original magnification 200×). C. This area of the lesion exhibits extensive areas of tubular and atubular dentin tissue within the nests of odontogenic epithelium (hematoxylin and eosin staining, original magnification 200×). D. In other regions, well-formed dental structures were noted, showing tubular dentin and enamel matrix (black arrow) (hematoxylin and eosin staining, original magnification 400×)

Source: the authors (SLMandic, Campinas, SP, Brazil).

The patient was followed up for 24 months postoperatively. He remained asymptomatic with no lesion recurrence (Figure 6). Soft tissue healing occurred uneventfully with bone remodeling.

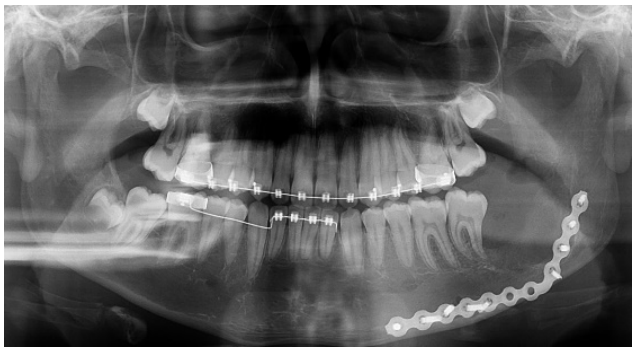

Figure 6 – Panoramic radiography showing after surgery there are no signs of recurrence

Source: the authors (SLMandic, Campinas, SP, Brazil).

Discussion

According to the 2005 WHO classification, AFO is a benign, mixed, and rare odontogenic neoplasm accounting for approximately 3% of odontogenic tumors[1,7]. The lesion affects persons aged below 20 years[4]. Buchner *et al.*[8] (2013) reported that most patients were aged between 5 and 14 years at the first diagnosis. The WHO classification describes the age range of the patient population as 8-12 years[1]. The literature shows a slightly higher prevalence among boys, although the WHO classification indicates no such gender-based predisposition[1,4,8]. In the present case, a 14-year-old boy presented with asymptomatic edema of rigid consistency

on palpation in the left and posterior region of the mandible and mild facial asymmetry. These clinical features are more common in the posterior region of the mandible, with asymptomatic and progressive growth; larger lesions can cause bone expansion[4,8].

AFO is often asymptomatic. The detection of AFO lesions is possible generally upon routine radiographic examination or during the investigation of a missing tooth[7,9]. Radiographic images of AFO lesions often show unilocular masses, although they can be multilocular and associated with an unerupted tooth, which may be dislocated[6]. The internal structure is often mixed with a radiolucent area adjacent to the well-defined margin. Typically, the lesion is mostly corticalized and may exhibit radiopacity that varies in shape and size internally. Some AFO lesions may present with a small amount of enamel matrix and dentin, displaying a radiolucent radiographic image[6-8]. These characteristics were consistent with the imaging findings in this case, which included a well-defined unilocular lesion, the presence of radiopaque material in the internal structure, and involvement of the unerupted third molar.

Careful evaluation of the clinical and radiographic imaging characteristics is crucial in the differential diagnosis of AFO, including differentiating from other odontogenic lesions such as AF, odontoma, calcifying odontogenic cysts, adenomatoid odontogenic tumors, and calcifying epithelial odontogenic tumors[10-12]. However, considering the location of the lesion in the posterior of the mandible and the posterior of the maxilla, the younger age at presentation, and the radiographic aspect of mixed density, some of these lesions could be excluded[1,6,10-12]. Therefore, the diagnostic hypothesis for our patient was AFO.

AFO is defined as a neoplasm composed of cords and islands of odontogenic epithelium included in the ectomesenchymal tissue with cells similar to the dental papilla, and with the presence of mineralized tissues, such as enamel matrix and dentin[1], characteristics found in the histopathological examination of the present case. According to the 2017 WHO classification, AFO and AFD were considered to have similar histological characteristics as those of developing odontomas. The analysis of the literature revealed that lesions may develop neoplastic characteristics and occur at ages not associated with the incidence of hamartomas[2,3]. AFO lesions appear in patients aged ≤20 years; the specific age in this range is an important factor in the differential diagnosis of the condition[6,13]. Slootweg[14] (1981) evaluated patients with AFO and reported a mean age of 8.1 years, while Buchner *et al*.[8] (2013) observed that the mean age was 9.6 years. However, Buchner and Vered[15] (2013) reported a mean age of 14.9 years in patients with AF lesions. Thus, the incidence of AFO may be associated with a lower mean age than that of the histologically primitive AF lesion; therefore, only few AFO lesions develop from AF injuries that later become odontomas[4,14,15].

Larger lesions cause cortical bone expansion, which may undergo perforations[7-10]. Thus, an AFO may have a higher growth potential that differs from that of a developing hamartoma, while other lesions are small and without bone expansion, and may represent developing odontomas[8,16,17]; AFOs may undergo malignant transformation, which does not occur in hamartomas[8,9]. Soluk-Tekkesin and Vered[18] found the areas under the curve (AUCs) of age and lesion size were significant for AFO and odontoma lesions, with AFO lesions being younger and larger in size. Future studies should investigate molecular and genetic specifications to improve our understanding of the pathogenesis of AFO lesions and odontomas.

Generally, AFO is treated conservatively, by extracting the impacted tooth and the involved lesion[9], although more extensive lesions may require wider surgical margins[8,10]. The prognosis is favorable as AFOs are less aggressive lesions with lower recurrence rates than AF[7-10]. Only a few studies described cases involving the malignant transformation of AFO into ameloblastic fibrosarcoma[6,7]. Hence, we proposed surgical with removal of the impacted third molar as the requisite treatment considering the large-sized lesion and bone involvement.

Conclusion

In conclusion, the clinical, radiographic, and histological aspects of AFO require accurate evaluation for a correct diagnosis and treatment. The proposed treatment was effective in our patient, and during the 24-month postoperative follow-up, no recurrence was noted.

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Ethical approval

This case report was approved by the Human Research Ethics Committee of São Leopoldo Mandic Dental School and Research Center (protocol CAAE nº 37173220.6.0000.5374), and patient anonymity has been ensured.

Resumo

Objetivo: o presente relato descreve as características clínicas, radiográficas e histopatológicas de uma lesão de fibro-odontoma ameloblástico (FOA). Relato de caso: relatamos o caso clínico de um menino de 14 anos com edema assintomático. A radiografia panorâmica detectou lesão unilocular com margens definidas e localizada na região posterior da mandíbula. A estrutura interna da lesão apresentava vários graus de radiopacidade com envolvimento do terceiro molar. A tomografia computadorizada de feixe cônico revelou as corticais ósseas vestibular e lingual expandidas, perfuração da cortical óssea lingual e deslocamento do canal mandibular. FOA foi a hipótese diagnóstica com base nas características radiográficas e clínicas. A excisão total foi realizada e examinada histologicamente, confirmando o diagnóstico de FOA. Nenhuma recorrência ocorreu durante um período de acompanhamento de 24 meses. Considerações finais: a avaliação das características clínicas, radiográficas e histopatológicas contribuíram para um diagnóstico correto e o tratamento adequado para o caso de FOA, uma vez que a lesão é frequentemente assintomática.

Palavras-chave: tumores odontogênicos; radiografia panorâmica; tomografia computadorizada de feixe cônico.

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