Ameloblastic Fibroma: A Case Report of a Rare Presentation Illustrating a Diagnostic Challenge

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Abstract

This case report emphasizes the diagnostic role of cone-beam computed tomography (CBCT) in identifying a rare pediatric case of ameloblastic fibroma (AF), comprising approximately 2% of odontogenic tumors. The 10-year-old patient initially presented with painless left mandibular posterior gingival swelling, resembling radicular or dentigerous cysts in clinical and conventional radiographic assessments. However, CBCT revealed a distinct periapical radiolucency linked to deciduous molar roots and an unerupted premolar, exposing mandibular lingual plate thinning and buccal plate disruption. The lesion extended beyond the unerupted premolar's enamel-cementum junction, suggesting an odontogenic tumor. Subsequent biopsy and histopathological examination confirmed AF. CBCT played a pivotal role in evaluating the lesion's interaction with surrounding bone structures, guiding treatment decisions, and contributing to an accurate diagnosis. Successful lesion excision, deciduous tooth extraction, and a disease-free year-long follow-up underscore the significance of enhancing diagnostic precision and managing uncommon odontogenic pathologies like AF.

Keywords: *Cone-beam computed tomography, differential diagnosis, odontogenic cysts, odontogenic tumors*

Introduction

Ameloblastic fibroma (AF) is a rare odontogenic tumor composed of ectomesenchymal tissue and odontogenic epithelial components. It represents approximately 2% of all odontogenic tumors, with around 279 documented cases in the literature. AF typically presents as an asymptomatic, well-defined, round osteolytic lesion in the mandibular region, primarily affecting young patients with an average age of 15.2 years and a slight male predominance.[1]

AF can exhibit similarities with various odontogenic cysts and tumors, including inflammatory cysts like radicular cysts (RC), developmental cysts such as dentigerous cysts (DC) and odontogenic keratocysts, or even odontogenic tumors like ameloblastoma, particularly.[2]

This case report presents a pediatric case of AF that posed a differential diagnosis challenge due to its radiographic similarities with both a radicular cyst and a dentigerous cyst. The report aims to discuss the differential diagnosis for this unique presentation and the contribution of cone-beam computed tomography (CBCT) in the diagnosis of a rare odontogenic tumor.

Case Report

Informed consent was obtained from the parents of the patient before the publication of this report. The complete clinical timeline is presented in Figure 1. A 10-year-old boy referred to the dental clinic with a painless swelling in the left mandibular posterior gingiva of unknown duration. There was no history of trauma, allergies, or recent systemic infection. The extraoral examination revealed no noticeable expansion or color changes. Intraoral examination revealed a firm, asymptomatic, normal-colored slight expansion on the buccal and lingual aspects of the cortical plates related to the left second deciduous molar. The tooth appeared healthy, without mobility or symptoms upon percussion. A discrete papule resembling a sinus tract was noted in the attached gingiva.

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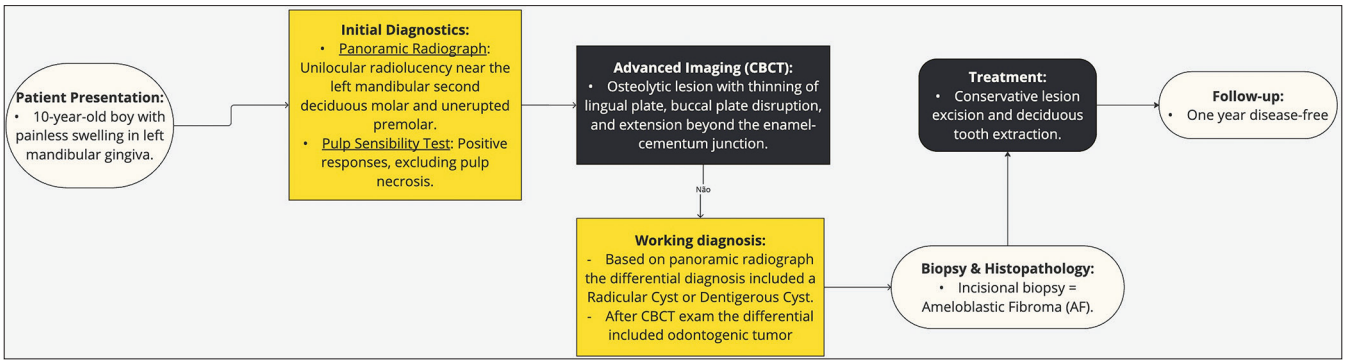

Figure 1: Clinical presentation timeline

A panoramic radiograph showed a well-defined periapical unilocular radiolucency associated with the roots of the left mandibular second deciduous molar and the coronal portion of the unerupted second premolar. The osteolytic lesion was associated with the resorption of the deciduous tooth roots and the mesial root of the first mandibular permanent molar [Figure 2a].

Based on the clinical and radiographic findings, the initial differential diagnosis included a RC or DC. To investigate the possibility of inflammation, a pulp sensibility test using tetrafluoroethane spray was performed, yielding positive responses in all teeth related to the radiolucent lesion.

CBCT was conducted to analyze the lesion's relationship with adjacent bone structures and infer its behavior. The CBCT scan revealed an osteolytic lesion associated with thinning of the lingual plate and disruption of the buccal plate of the left mandible body, possibly in the region where the presumed "sinus tract" was observed. In addition, the lesion extended beyond the enamel-cementum junction of the unerupted premolar [Figure 2b-e].

A hypothesis of an odontogenic tumor was proposed. A needle aspiration yielded no fluid, and an incisional biopsy was performed under local anesthesia. Histopathological examination revealed connective stromal tissue resembling a primitive dental papilla permeated by strands and islands of apparently inactive odontogenic epithelium [Figure 3a-d], confirming the diagnosis of AF. The lesion was conservatively excised, and the associated deciduous tooth was extracted. The patient has remained disease-free during rigorous follow-up for the past year [Figure 3e and f].

Discussion

AF is an odontogenic tumor primarily affecting the pediatric population.[1] It should be considered when an unknown radiolucency is observed in the posterior mandible of young patients, particularly when not associated with dental pulp necrosis.[3] The tumor typically presents as a painless expansion associated with a variable-sized unilocular radiolucency, although larger lesions may display a multilocular appearance.[1] Cortical bone perforation and tooth resorption are less frequent.[2]

AF accounts for 1.6% of all odontogenic tumors.[3] However, it may have a higher prevalence in the pediatric population alone, highlighting the need for further research to improve understanding and diagnosis.

Conservative surgical procedures such as excision or curettage are the primary treatment options for AF. Recurrent or larger tumors may require more radical excisions.[4] The recurrence rate is approximately 33%, with conservative treatments associated with a higher recurrence rate. Malignant transformation has been reported in 11% of recurrent cases, underscoring the importance of long-term follow-up.[4]

When encountering radiolucent lesions associated with a tooth, odontogenic cysts should be considered in the differential diagnosis. In this case, a RC was initially considered due to the radiolucency associated with the apex of a tooth root, suggesting an osteolytic lesion resulting from infection or pulpal necrosis.[5] However, RC is typically related to permanent teeth with dental caries or large restorations, which contrasts with the current presentation.

A DC was also considered, as the radiolucency was associated with the crown of an unerupted permanent tooth, possibly caused by an inflammatory process extending through the apex of the overlying deciduous tooth. In addition, it is a common lesion in the pediatric population.[2,6] However, the deciduous tooth did not respond positively to the sensibility test, and the CBCT exams showed that the attachment of the cystic lesion extended beyond the cement-enamel junction, excluding the possibility of a DC.

A key strength of this case report is the pivotal role that CBCT played in the diagnosis, as it was essential for analyzing the lesion's relationship with adjacent bone structures and for inferring its behavior. The CBCT scan revealed an osteolytic lesion associated with thinning of the lingual plate and disruption of the buccal plate of the left mandible body, potentially in the area where the presumed "sinus tract" was identified. This precise imaging allowed for the formulation of a hypothesis suggesting an odontogenic tumor. Another strength is the use of biopsy

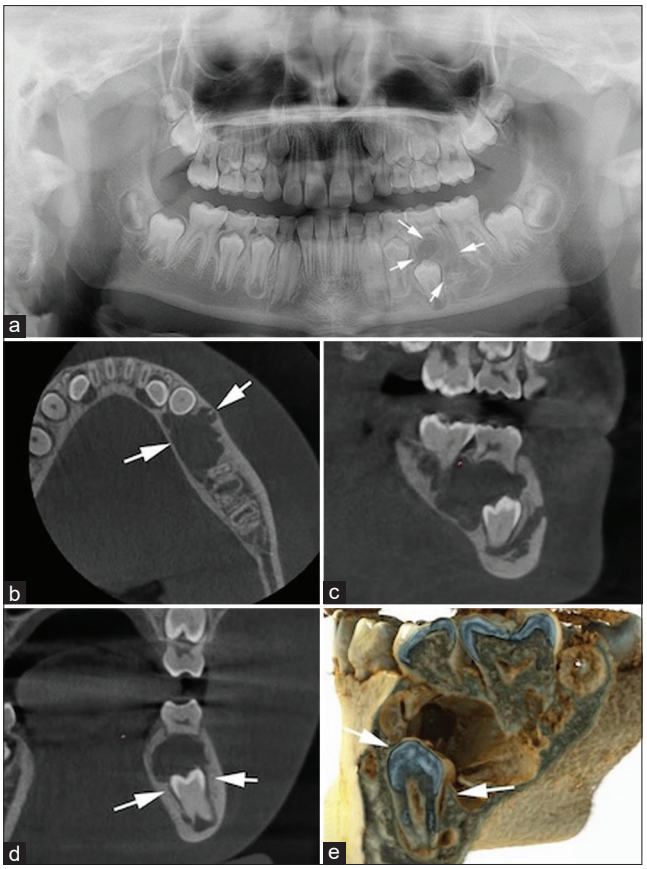

Figure 2: (a) Panoramic radiograph image showing a well-defined periapical unilocular radiolucency (indicated by arrows) associated with the roots of the left mandibular second deciduous molar and the coronal portion of the unerupted second premolar. Cone-beam computed tomography scans presenting in (b) axial reconstruction an osteolytic lesion with thinning of the lingual plate and disruption of the buccal plate of the left mandible body (indicated by arrows). (c) Sagittal reconstruction showing the osteolytic lesion associated with resorption of the deciduous tooth root. (d and e) Coronal and three-dimensional reconstructions (realistic filter – e-Vol DX, CDT Software, São José, SP, Brazil) exhibiting the lesion extension beyond the enamel-cementum junction of the unerupted tooth (indicated by arrows)

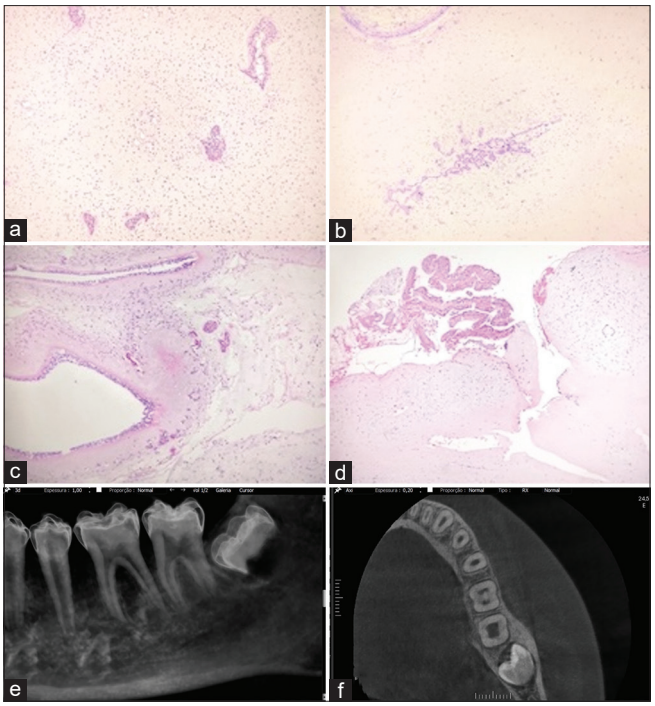

Figure 3: Microscopic aspects of ameloblastic fibroma. (a and b), Cords and small islands of odontogenic epithelium in a loose mesenchymal tissue. (c) Ameloblastic differentiation on the periphery of some nests. (d) Reduced enamel epithelium of the associated tooth. (H and E, ×200 original magnification). Cone-beam computed tomography scans in the sagittal (e) and axial (f) reconstructions demonstrate no evidence of disease

and histopathological examination, which were crucial in providing a definitive diagnosis of AF and determining the need for long-term clinical follow-up due to the possibility of malignant transformation.[7,8]

This case report demonstrates the clinical management and differential diagnosis of a rare case of AF in a pediatric patient. However, there are some limitations. While CBCT was crucial for the initial diagnosis, it cannot substitute for a biopsy and histopathological analysis to confirm the diagnosis. Furthermore, as this is a report of a single case of a rare condition, the findings may not be generalizable to all cases of AF.

From the patient's perspective, the experience was overall positive, with no pain from the initial swelling and minimal discomfort during diagnostic procedures, including CBCT and a biopsy under local anesthesia. Informed consent was obtained from the parents, and the diagnosis of AF, though rare, was managed with a conservative excision of the lesion and an associated tooth. The patient has remained disease free for a year, and regular follow-up has provided reassurance about long-term outcomes.

Overall, this case report highlights that AF should be considered in cases of unknown radiolucency in the posterior mandible of young patients, particularly when not associated with dental pulp necrosis; long-term follow-up is necessary to monitor the patient's condition and detect any potential recurrence or malignant changes; biopsy and histopathological exams are necessary for rule out malignancy.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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