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Case Report

Ameloblastic Fibroma Associated with an Impacted Mandibular Third Molar: A Case Report

Pryscilla Batista Leite – DDS. Oral and Maxillofacial Surgeon at the Pedro Ernesto's Hospital from the State University of Rio de Janeiro, Brazil. Mailing address: Boulevard 28 de setembro, 77 - Rio de Janeiro – RJ, Brazil. CEP 20.551-030. email: pryscilla_leite@hotmail.com  <https://orcid.org/0000-0003-4062-8153>

Vera Campos - DDS. MS. Assistant professor from the Department of Preventive and Community Dentistry at the State University of Rio de Janeiro's Dental School, Brazil. Mailing address: Boulevard 28 de setembro, nº 157, sala 226, Vila Isabel, Rio de Janeiro – RJ, Brazil. CEP 20.551-031. email: prof_vcampos@yahoo.com.br  <https://orcid.org/0000-0002-5355-6999>

Michele Machado Lenzi da Silva - DDS. MS. PhD. Adjunct professor from the Department of Preventive and Community Dentistry at the State University of Rio de Janeiro's Dental School, Brazil. Mailing address: Boulevard 28 de setembro, nº 157, sala 226, Vila Isabel, Rio de Janeiro – RJ, Brazil. CEP 20.551-031. email: mmlenzi82@gmail.com  <https://orcid.org/0000-0002-6837-0564>

Mirian de Waele Souchois de Marsillac - DDS. MS. PhD. Associate professor from the Department of Preventive and Community Dentistry at the State University of Rio de Janeiro's Dental School, Brazil. Mailing address: Boulevard 28 de setembro, nº 157, sala 226, Vila Isabel, Rio de Janeiro – RJ, Brazil. CEP 20.551-031. e-mail: mwsm36@gmail.com  <https://orcid.org/0000-0002-4452-7336>

Fábio Ramoa Pires - DDS, MS, PhD. Associate professor, Oral Pathology, State University of Rio de Janeiro's Dental School. Mailing address: Boulevard 28 de setembro, nº 157 - Vila Isabel, Rio de Janeiro – RJ, Brazil. CEP 20.551-031. e-mail: ramoaafop@yahoo.com  <https://orcid.org/0000-0003-0317-8878>

Corresponding author: Pryscilla Batista Leite

e-mail: pryscilla_leite@hotmail.com; Mailing address: Boulevard 28 de setembro, nº 62, sala 506 Vila Isabel, Rio de Janeiro – RJ, Brazil. CEP 20.551-031; Telephone: +55 (21) 993068585

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ABSTRACT

Ameloblastic fibroma (AF) is a rare benign mixed odontogenic tumor that affects the jaws, most commonly occurring in the posterior region of the mandible. It predominantly affects individuals during the first two decades of life and is more prevalent in males. Histopathologically, AF is characterized by islands and cords of odontogenic epithelium embedded within an ectomesenchymal stroma resembling the dental papilla and enamel organ. This article reports the case of a 12-year-old female patient with albinism in whom a routine radiographic examination revealed a well-defined unilocular radiolucent lesion in the posterior right region of the mandible associated with an impacted third molar. Clinical examination revealed no facial asymmetry or buccal cortical expansion. The lesion was treated by excisional biopsy and enucleation, with preservation of the impacted tooth. Histopathological analysis confirmed the diagnosis of ameloblastic fibroma. The patient remains under clinical and radiographic follow-up, demonstrating progressive bone regeneration and preservation of tooth 48, with no evidence of recurrence 17 months after surgery. This case highlights the value of routine radiographic examination and conservative surgical management in young patients, allowing lesion removal while preserving the associated tooth and with favorable bone

healing.

Keywords: Fibroma; Odontogenic Tumors; Oral Surgical Procedures; Mandibular Neoplasms; Radiography, Panoramic.

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INTRODUCTION

Ameloblastic fibroma (AF) is a rare benign mixed odontogenic tumor composed of both epithelial and mesenchymal components¹⁻⁴. It was first described in the literature by Kruse in 1891 and accounts for approximately 1.5% to 4.5% of all odontogenic tumors⁶.

According to the literature, AF predominantly affects patients during the first and second decades of life, with approximately 75% of cases diagnosed before the age of 20 years, characterizing it as a tumor of childhood and adolescence⁷. The lesion exhibits a male predilection, with an approximate male-to-female ratio of 1.4:1⁸.

AF most commonly occurs in the posterior segment of the mandible, with approximately 80% of cases located in the region of the second primary molar or first permanent molar⁷. Furthermore, approximately 75% of cases are associated with impacted teeth². Due to the absence of painful symptoms, AF is frequently diagnosed incidentally during routine radiographic examinations. Radiographically, it presents as a well-circumscribed radiolucent lesion, often surrounded by a radiopaque border, and may be associated with an impacted tooth, exhibiting either a unilocular or multilocular appearance.

Histopathologically, AF is characterized by the presence of islands and cords of odontogenic epithelium embedded within a loose and primitive connective tissue stroma resembling the dental papilla. Epithelial structures resembling the follicular stage of the developing enamel organ may also be observed¹⁰⁻¹².

The differential diagnosis of AF includes other odontogenic lesions that present as radiolucent images, particularly ameloblastoma, dentigerous cyst, and odontogenic keratocyst^{14,4}.

With regard to treatment, conservative management consisting of tumor enucleation and curettage is generally recommended for smaller lesions^{15,11}. For larger lesions, resection of the affected region may be indicated^{16,17}.

Although AF is an uncommon entity, it is clinically relevant because of its radiographic similarity to other odontogenic lesions and its potential for recurrence,

highlighting the importance of an accurate diagnosis and appropriate management. Therefore, the aim of this study is to report the case of a 12-year-old female patient with albinism diagnosed with AF in the posterior right region of the mandible associated with an impacted tooth 48 highlighting the conservative surgical approach, preservation of the involved tooth, and favorable bone regeneration during follow-up.

CASE REPORT

A 12-year-old female patient with albinism and no systemic comorbidities attended the Pediatric Dentistry Clinic at the State University of Rio de Janeiro (UERJ) for routine dental evaluation prior to orthodontic treatment.

Panoramic and lateral cephalometric radiographs revealed a well-defined unilocular radiolucent lesion with a sclerotic border associated with tooth 48 (Figures 1A and 1B). Intraoral clinical examination showed no swelling and the patient stated no local pain. Cone-beam computed tomography (CBCT) was subsequently requested and demonstrated that the lesion extended buccally and lingually in the mediolateral dimension without causing cortical bone expansion. In the anteroposterior dimension, the lesion extended from the mesial aspect of the mandibular right second molar to the inferior portion of the mandibular ramus (Figures 2). The initial diagnostic hypotheses were dentigerous cyst and odontogenic keratocyst, and the patient was referred to the Oral and Maxillofacial Surgery team for surgical management.



Figure 1. (A,B) Panoramic and lateral cephalometric radiographs showing a well-defined unilocular radiolucent lesion with a sclerotic border associated with tooth 48.

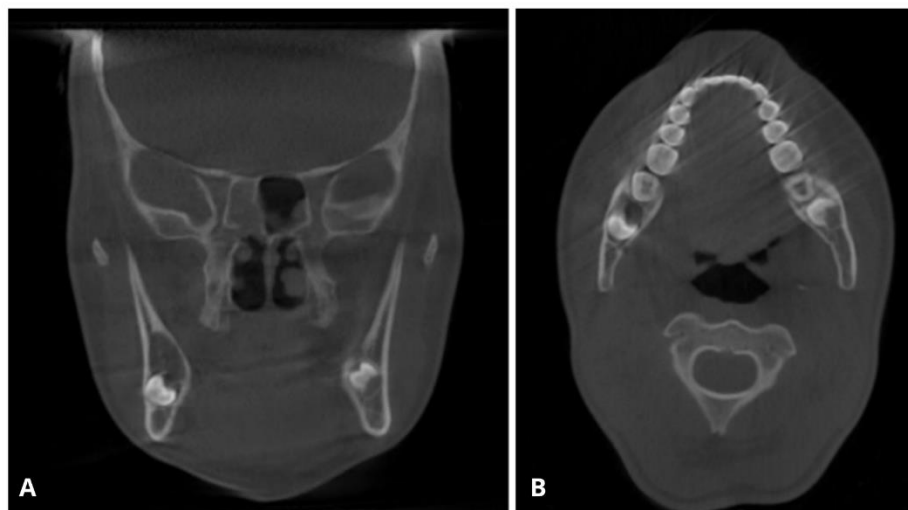


Figure 2. (A) Coronal CBCT section showing mediolateral extension of the lesion toward both the buccal and lingual aspects without cortical bone expansion. **(B)** Axial CBCT section demonstrating anteroposterior extension of the lesion from the mesial aspect of the mandibular right second molar to the inferior portion of the mandibular ramus.

Local anesthesia was administered to the inferior alveolar, lingual, and buccal nerves using 2% lidocaine with 1:100,000 epinephrine. Following mucoperiosteal flap elevation, a conservative osteotomy was performed to access the lesion. Enucleation was carried out while preserving tooth 48 due to its stage of development and potential for future eruption. Curettage of the surgical site was performed, followed by copious irrigation with 0.9% saline solution. The wound was closed with absorbable sutures (Vicryl® 3-0), and the surgical specimen was submitted for histopathological analysis.

The specimen underwent routine laboratory processing, and hematoxylin and eosin-stained sections revealed long, thin cords of odontogenic epithelium, frequently exhibiting anastomosis and composed of cuboidal to columnar cells. Small epithelial

islands resembling the follicular stage of enamel organ development were also observed. Peripheral columnar cells surrounded loosely arranged epithelial cells resembling the stellate reticulum. These epithelial structures were embedded within a loose connective tissue stroma composed of spindle-shaped and stellate cells, resembling the architecture of the dental papilla (Figure 3). Histopathological examination confirmed the diagnosis of ameloblastic fibroma. No cellular atypia, pleomorphism, or mitotic figures were observed, supporting the benign nature of the lesion.

The postoperative period had no recurrence, and the patient was maintained under clinical and radiographic follow-up. A panoramic radiograph obtained 17 months after surgery demonstrated substantial bone regeneration and continued development of tooth 48 (Figure 4). The patient remains under follow-up, with no pain, neurosensory disturbances, or signs of recurrence.

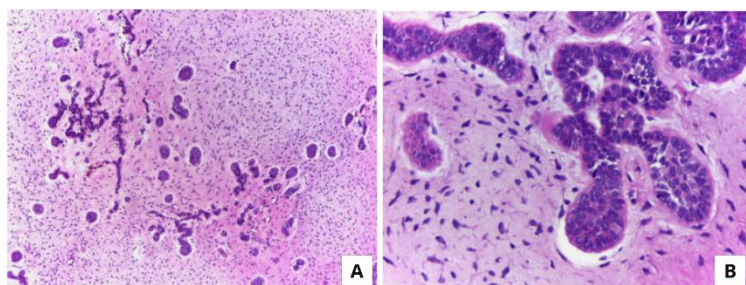


Figure 3. (A) Proliferation of epithelial islands and cords embedded within a hypercellular loose connective tissue stroma (H&E stain, original magnification $\times 100$). **(B)** Higher magnification showing epithelial proliferation with peripheral columnar cells and central cells resembling the stellate reticulum, surrounded by mesenchymal cells with spindle-shaped and stellate nuclei (H&E stain, original magnification $\times 400$).

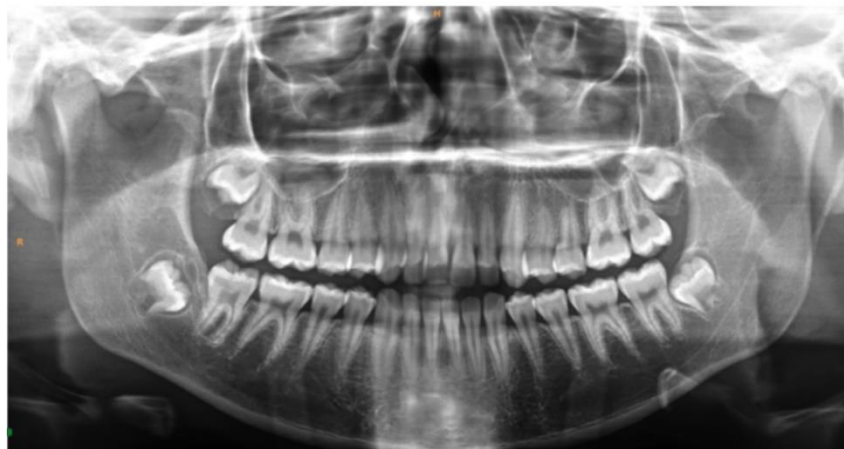


Figure 4 - Panoramic radiograph obtained 17 months after lesion removal, showing eruption and continued development of tooth 48 (Nolla stage 6) associated with bone regeneration in the affected area.

DISCUSSION

Ameloblastic fibroma (AF), first described by Kruse⁵ is a rare benign mixed odontogenic tumor composed of epithelial and mesenchymal components^{5,30,18}. It most commonly occurs in the posterior region of the mandible and is frequently associated with impacted teeth².

The case presented in this report involved a 12-year-old female patient. This finding is consistent with the age range most reported in the dental literature^{19,7}, which indicates a mean patient age of approximately 14 years^{21,11}. Although AF exhibits a male predominance, with a male-to-female ratio of approximately 1.4:1⁷, the present case occurred in a female patient.

An additional noteworthy feature of the present case is the coexistence of ameloblastic fibroma and albinism. Although no established epidemiological or pathogenic association between these conditions has been demonstrated, this finding

may be of biological interest. Albinism is characterized by alterations in melanogenesis, particularly involving the tyrosinase-dependent pathway, whereas melanocytes are neural crest-derived cells that may be identified, albeit rarely, within odontogenic tissues and tumors^{23,29}. Pigmentation has exceptionally been described in odontogenic lesions, including ameloblastic fibroma, suggesting that melanocytic cells may occasionally be retained or incorporated into odontogenic neoplasms¹⁸. Therefore, the coexistence of albinism and ameloblastic fibroma in the present patient raises a hypothesis regarding whether alterations in melanocytic biology could influence the odontogenic microenvironment²³. However, this hypothesis remains speculative, and the available literature does not support a causal relationship between albinism and the development of ameloblastic fibroma. Further cases and molecular studies would be necessary to determine whether this association is coincidental or reflects an underlying biological relationship.

Radiographically, AF usually presents as a well-defined radiolucent lesion, frequently surrounded by a sclerotic border (94%), and may appear as a small unilocular lesion (56%) or as a larger multilocular lesion (44%)²². The differential diagnosis includes dentigerous cyst, ameloblastoma, odontogenic keratocyst, and odontogenic myxoma¹⁵. In the present case, the association with an impacted tooth initially suggested the diagnosis of a dentigerous cyst. Although conventional radiographic examinations play a fundamental role in diagnosis^{24,6}, computed tomography is particularly useful for determining lesion extent and planning surgical treatment^{7,6,15}. In the present case, a lateral cephalometric radiograph was additionally obtained as part of the orthodontic treatment planning.

Within the differential diagnosis of AF, it is important to exclude ameloblastoma and dentigerous cyst^{14,4,30}. In the present case, ameloblastoma was considered less likely based on the patient's age, well-defined unilocular appearance, and absence of characteristic histopathological features. A dentigerous cyst was also considered but the clinical, radiographic, and histopathological findings supported the diagnosis of AF.

Histopathological evaluation should carefully assess the presence of cellular and nuclear pleomorphism, typical and atypical mitotic figures, and areas of necrosis in order to exclude ameloblastic fibrosarcoma¹⁰⁻¹¹. None of these features were identified in the present case, supporting the diagnosis of AF. Histopathological findings in hematoxylin and eosin-stained showed features that characterized an AF specimen, as described by

previous authors¹⁰⁻¹².

In the present case, the lesion was treated by enucleation and curettage, while preserving the impacted tooth (48) because of its favorable position and potential for future eruption. The favorable postoperative outcome, associated with bone regeneration and continued development of tooth 48, supports the conservative treatment approach adopted in this case. This surgical approach was based on the existing literature^{19,25,27}, which indicates that this is the treatment of choice for AF. This lesion is generally considered a non-aggressive neoplasm, complete surgical removal is associated with favorable outcomes; however, recurrence rates ranging from 18% to 43.5% have been reported following conservative enucleation^{28,27}. Recurrence is usually attributed to incomplete tumor removal and the persistence of residual neoplastic tissue⁶.

Even though ameloblastic fibroma is a benign tumor, malignant transformation into ameloblastic fibrosarcoma has already been reported, especially in recurrent lesions. There is histological evidence of a pre-existing ameloblastic fibroma in 44% of ameloblastic fibrosarcoma cases, emphasizing the need for long-term follow-up.¹³ It's reasonable to consider that the locally aggressive behavior of ameloblastic fibrosarcoma has a reported recurrence in 37% of cases and mortality in 19.3% of patients⁹.

After 17 months of follow-up, the patient remains asymptomatic, with radiographic evidence of bone regeneration, preservation of the impacted tooth (48), and no signs of recurrence. Nevertheless, continued long-term follow-up remains necessary because recurrence has been reported after surgical treatment of AF. Long-term clinical and radiographic follow-up is recommended to ensure successful treatment outcomes and early detection of recurrence^{6,4}.

CONCLUSION

Ameloblastic fibroma is a rare benign mixed odontogenic tumor with a favorable prognosis when treated appropriately. Accurate clinical, radiographic, and histopathological evaluation is essential for establishing the diagnosis and guiding treatment. In the present case, conservative surgical management with preservation of

the impacted tooth resulted in satisfactory bone regeneration and no evidence of recurrence after 17 months of follow-up. Preservation of the associated unerupted tooth was advantageous, given its stage of tooth development and potential for spontaneous eruption following lesion removal. Long-term clinical and radiographic monitoring remains essential to ensure treatment success and allow early detection of recurrence.

Author contributions:

- Pryscilla Batista Leite have made substantial contributions to acquisition of data; analysis and interpretation of data; been involved in drafting the manuscript and revising it critically for important intellectual content; and given final approval of the version to be published; and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.
- Michele Machado Lenzi da Silva, Vera Campos, Fábio Ramoa Pires and Mirian de Waele Souchois de Marsillac have made substantial contributions to conception and design; acquisition of data; analysis and interpretation of data; and been involved in drafting the manuscript and revising it critically for important intellectual content; and given final approval of the version to be published; and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

CONFLICT OF INTEREST STATEMENT

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Competing interests: The authors have no relevant financial or non-financial interests to disclose.

Ethics approval: This study is part of the research project titled "Hereditary/genetic, congenital, and acquired oral alterations in children and/or adolescents treated at the Pediatric Dentistry Clinic of FOUERJ: a prospective cohort," which was approved by the Human Research Ethics Committee of the State University of Rio de Janeiro (UERJ), Protocol No. 352 ESPO.

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