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Odontogenic Puzzle in a 3-Year-Old: Unveiling an Ameloblastic Fibroma

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Abstract

Ameloblastic fibroma (AF) is a rare mixed odontogenic tumor, accounting for 1.5%–4.5% of all odontogenic tumors. It commonly arises during the first and second decades of life and appears radiographically as a well-defined unilocular or multilocular radiolucency. Although considered benign, AF has potential for recurrence and malignant transformation into ameloblastic fibrosarcoma, necessitating careful management and long-term follow-up. A conservative approach, enucleation with curettage, and long-term follow-up are absolutely necessary for any recurrence or change to fibrosarcoma. We report a case of AF in a 10-year- old male patient who presented with a chief complaint of swelling in the right mandibular posterior region. Enucleation and curettage were done under general anesthesia.

Keywords: Ameloblastic Fibroma, Ameloblastic fibrosarcoma, Enucleation and Curettage

Introduction

Ameloblastic fibroma is a rare mixed odontogenic benign tumor that can occur in either the mandible or maxilla, although it is most commonly found in the posterior region of the mandible and is often associated with unerupted teeth. The lesion was first described by Kruse in 1892, and later characterized as a benign neoplasm by Thoma and Goldman in 1946. In 1992, the World Health Organization included it in the classification of mixed odontogenic tumors.

The frequency of occurrence of ameloblastic fibroma ranges between 1.5% and 4.5% of all odontogenic tumors. According to Reichart and Philipsen, there is a male-to-female predilection of 1.4:1, with the average age of occurrence around 14 years, although it can range from 2 to 62 years. It is a slowly developing tumor typically located in the posterior mandibular region and is mostly associated with an impacted tooth. In cases where the tumor is near the surface of bone, slight buccal and lingual cortical plate expansion may be observed.

Radiographically, ameloblastic fibromas appear as well-defined unilocular or multilocular radiolucencies with sclerotic radiopaque borders. Unilocular lesions are often asymptomatic and discovered incidentally during routine dental examinations, while multilocular lesions commonly present as jaw swelling. Of particular clinical significance is the potential for ameloblastic fibroma to transform into ameloblastic fibrosarcoma in about 45% of cases.

Histologically, ameloblastic fibromas consist of both epithelial and connective tissue components. The connective tissue resembles dental papilla and contains spindled and angular cells with delicate collagen, giving it a myxomatous appearance. Variants of ameloblastic fibroma include granular cell ameloblastic fibroma and papilliferous ameloblastic fibroma.

The conventional treatment involves enucleation with curettage of the adjacent bone, accompanied by extraction of the affected tooth. Long-term follow-up is essential to monitor for any recurrence.

Case Report

A 3-year-old male patient reported to our department with the chief complaint of a hard, painless swelling on the right side of his cheek, present for one month. The swelling had an insidious onset, with no history of trauma. Medical and family histories were noncontributory.

Extraoral examination revealed a diffuse swelling on the right lower third of the face, measuring approximately 4 cm × 2 cm, extending from 2 cm below the ala-tragus line to the lower border of the mandible, and from the corner of the mouth to 1 cm anterior to the angle of the mandible. On palpation, the swelling was firm, non-fluctuant, noncompressible, and nontender. No ulceration or drainage was observed.

Intraoral examination showed a swelling extending from the distal of the canine to the first molar region. The overlying mucosa was similar to the surrounding mucosa, and the swelling measured approximately 2 cm × 3 cm, with obliteration of the buccal vestibule. On palpation, the swelling was bony hard, with mild tenderness.

Imaging studies revealed a well-defined unilocular radiolucency with sclerotic borders, extending from the distal of the canine to the first molar region, associated with an impacted tooth. Radiographically, the lesion resembled a unilocular radiolucency with smooth, well- demarcated borders, often misdiagnosed as a dentigerous cyst. Intraorally, cortical expansion of the affected bone was evident, demonstrated by an increased mesiodistal distance between the mandibular first molar and the adjacent mandibular deciduous second molar.

Based on the clinical and radiological findings, a provisional diagnosis of dentigerous cyst was made, and the patient was planned for marsupialization. After removing a portion of the cystic lining, a sample was sent for histopathological examination, which revealed the diagnosis of ameloblastic fibroma.

Subsequently, the patient underwent surgical enucleation followed by chemical cauterization of the lesion.

Macroscopically, the excised tissue was brownish, soft in consistency, measuring approximately:

(A) 2.5 cm × 1.8 cm × 1.5 cm
(B) 1.5 cm × 1.1 cm × 1.0 cm.

Microscopic examination showed ameloblastic islands with keratinization, focal areas containing compressed epithelial strands and cords, and epithelial islands lined by elongated ameloblast-like cells. Some cells exhibited clear cytoplasm, resembling clear cells. The connective tissue stroma was moderately collagenous with chronic inflammatory infiltrates, mainly lymphocytes and plasma cells. Blood vessels lined by endothelial cells were also present

Discussion

Ameloblastic fibroma (AF) was first described by Kruse in 1892, later characterized as a benign neoplasm by Thoma and Goldman in 1946, and in 1992, it was classified by the World Health Organization as a mixed odontogenic neoplasm. AF is a true mixed odontogenic tumor comprising both epithelial and mesenchymal components, primarily occurring in young patients during the first or second decades of life. It is predominantly found in the posterior region of the mandible and is more common in males than females, with a male-to- female predilection of 1.4:1{1-4}

Histologically, the epithelial and connective tissue components of AF reflect the cap and bell stages of odontogenesis. The frequency of occurrence ranges from 1.5% to 4.5% of odontogenic tumors, and the average age of presentation is around 14 years, with a broad range from 2 to 62 years. AF is a slow-growing tumor that is commonly associated with an impacted tooth, and in superficial cases, slight buccal and lingual cortical expansion may be observed.{5,6}

Several histological variants of AF have been described, including:

Volume 8, Issue 5; September-October 2025; Page No 501-506
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1. Granular cell ameloblastic fibroma – characterized by granular cells in the ectomesenchyme.
2. Papilliferous ameloblastic fibroma – marked by prominent epithelial proliferation with a plexiform pattern.
3. Ameloblastoma associated with ameloblastic fibroma.
4. Cystic ameloblastic fibroma.{7}

The pattern and direction of epithelial follicle enlargement are influenced by the density of surrounding collagen fibers, growing in planes of least resistance when collagen is dense.

In our case, there was no evidence of hard tissue formation, effectively ruling out ameloblastic fibroodontoma or fibrodentinoma. No atypical features or mitotic activity were observed. Microscopically, the tumor consisted of strands and islands of odontogenic epithelium embedded in a primitive connective tissue stroma resembling dental papilla, with small islands resembling the follicular stage of enamel organ development. Recurrent cases of AF have been reported to show dentin formation, with or without enamel structures, and in some cases, may differentiate into odontomas.

The presence of mitotic figures should prompt consideration of differential diagnoses such as ameloblastic fibrosarcoma, where malignant transformation involves prominent changes in the mesenchymal component and loss of odontogenic epithelium.

Regarding treatment, approaches vary between conservative excision and aggressive resection. Philipsen et al. proposed that the typically innocuous behavior of AF does not justify aggressive initial treatment, advocating instead for meticulous surgical enucleation with close clinical follow-up.

Conservative Approach – Enucleation And Curettage:

This is the most commonly employed method, especially in young patients. Enucleation involves complete removal of the tumor along with curettage of the surrounding bone to remove residual tumor cells. This approach is favored to preserve surrounding structures and reduce morbidity, particularly in growing children where aggressive resection may impair jaw development, function, and aesthetics. Studies show that conservative treatment has acceptable success rates, especially when close clinical and radiographic follow-up is maintained.

Aggressive Approach – Resection With Margins:

In cases where the tumor is large, recurrent, or exhibits aggressive histopathological features, some authors recommend marginal or segmental resection of the affected jaw. This ensures complete removal of the tumor and reduces recurrence rates but comes at the cost of greater surgical morbidity. Resection is often reserved for recurrent cases or when histopathology reveals atypical or aggressive features.

Nevertheless, malignant transformation into ameloblastic fibrosarcoma, although rare, is well documented. Additionally, recurrence rates reported in the literature are notable: 43.5% by Trodahl et al. and 18.3% by Zallen et al., highlighting the need for careful management.

In the present case, based on the biopsy report confirming AF, an CYSTIC ENUCLEATION FOLLOWED BY CHEMICAL CAUTERIZATION was performed under general anesthesia.

Malignant Transformation Risk

Although rare, ameloblastic fibroma can undergo malignant transformation into ameloblastic fibrosarcoma, particularly in recurrent cases. This risk, though low, emphasizes the importance of thorough histopathological evaluation and long-term surveillance. If malignant transformation is suspected or confirmed, more aggressive surgical management, possibly with adjuvant therapy, is warranted.

Proliferation markers such as MIB-1, a monoclonal antibody against nuclear proliferation- associated antigens, have been studied in AF, with labeling indices ranging from 2.9% to 7.5% in the epithelial component and 1.5% to 13.5% in the mesenchymal component. These indices tend to be higher in recurrent AF and in cases that have undergone malignant transformation.{10}

Conclusion

Ameloblastic fibroma is a benign odontogenic mixed tumor with a generally excellent prognosis. The standard treatment is typically conservative, involving enucleation and curettage of the surrounding tissue, usually under local or general anesthesia depending on the case.The treatment of ameloblastic fibroma should balance effective tumor removal with preservation of

Volume 8, Issue 5; September-October 2025; Page No 501-506
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function, particularly in pediatric patients. A conservative surgical approach (enucleation and curettage) remains the first-line treatment, provided close postoperative monitoring is maintained. In recurrent or aggressive cases, resection with clear margins may be necessary to prevent further recurrence or malignant transformation. Given the rare and unpredictable nature of AF, a personalized, multidisciplinary treatment plan is recommended, ensuring optimal outcomes with minimal long-term complications Due to its relatively high risk of recurrence, long-term follow-up is essential following surgical removal to monitor for recurrence or potential malignant transformation.

Ethical Considerations

This case report was conducted in accordance with the ethical standards outlined in the Declaration of Helsinki. Informed consent was obtained from the child's legal guardians prior to the collection of clinical data, radiographic images, and publication of anonymized photographs and relevant details. The patient's identity has been fully protected, with no personal identifiers disclosed to maintain confidentiality. The decision to publish this case was made with the primary intention of contributing to scientific knowledge and advancing understanding of rare odontogenic tumors in pediatric patients, while prioritizing the best interests and privacy of the patient. The study did not involve any experimental treatment, and all clinical management adhered to established medical and ethical guidelines.

Institutional ethical approval was obtained where necessary, and the family was provided full transparency regarding the purpose and implications of the report.

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Preoperative Photographs

Volume 8, Issue 5; September-October 2025; Page No 501-506
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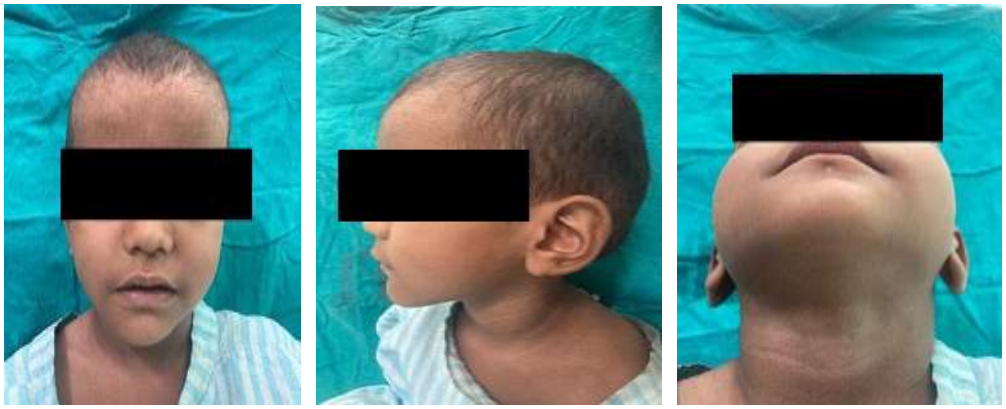
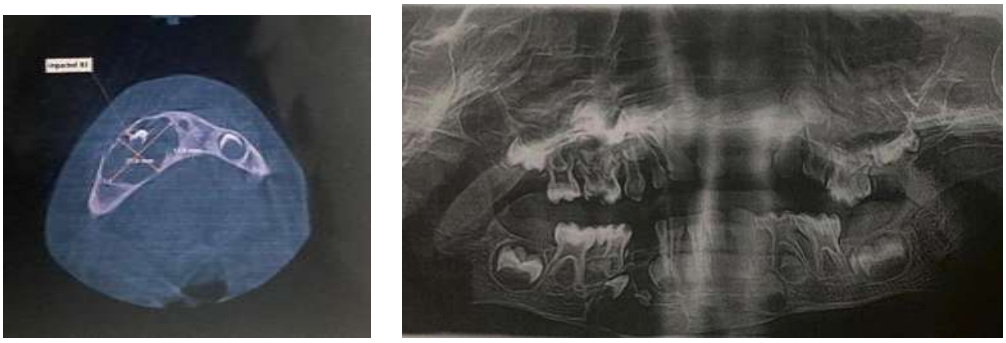
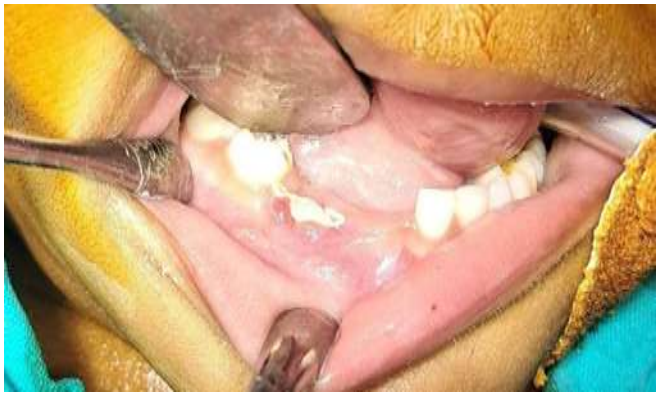
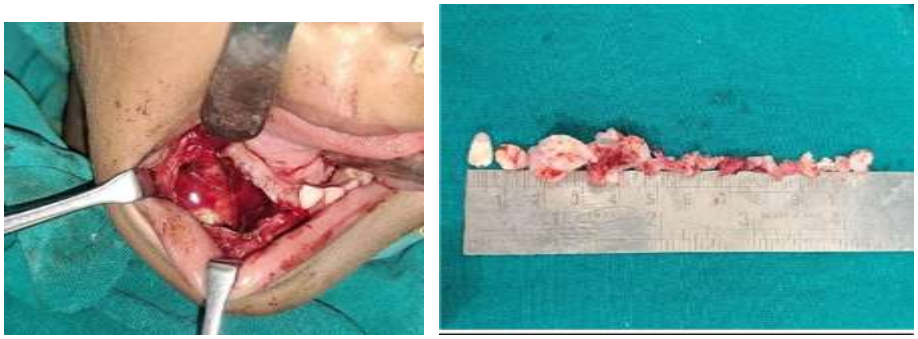






Intraoperative Photograph



Volume 8, Issue 5; September-October 2025; Page No 501-506

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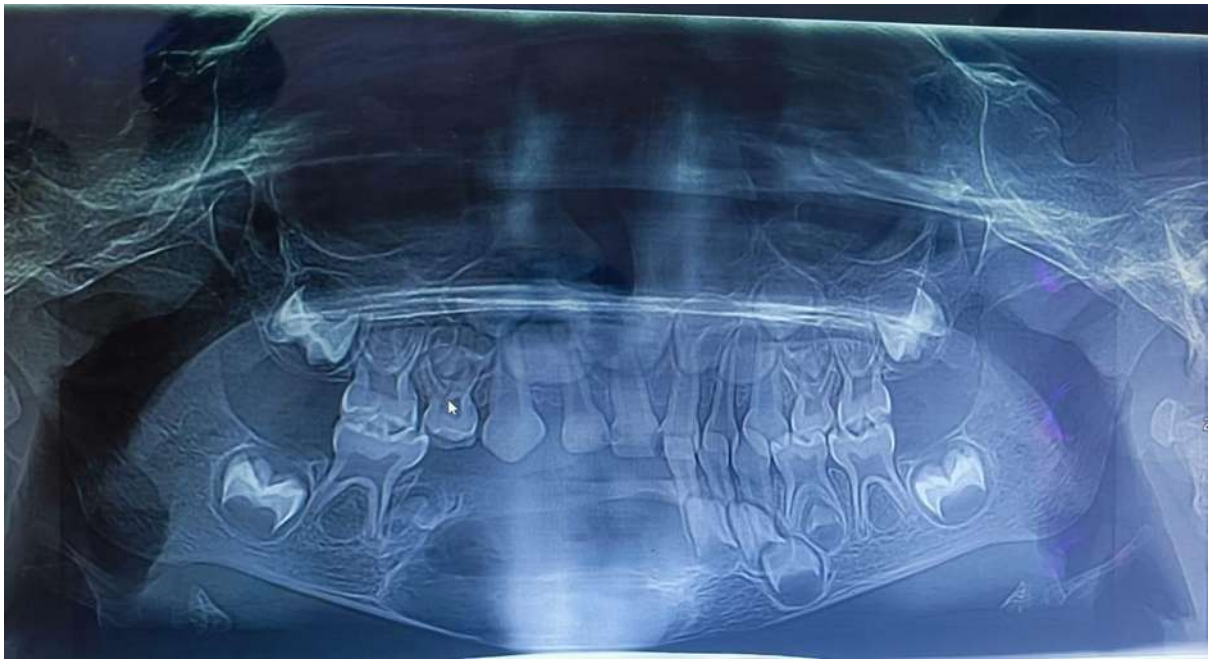
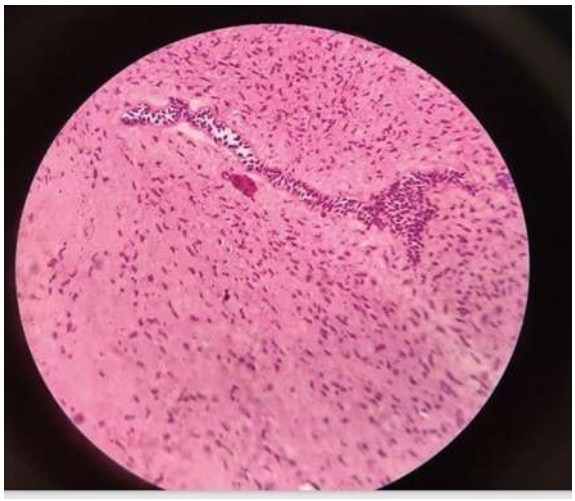
Postopertive Photograph



Histopathological Phot Ograph



Volume 8, Issue 5; September-October 2025; Page No 501-506

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