

Case Report

A growing jaw meets a growing tumor: Pediatric ameloblastic fibroma

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Abstract

Ameloblastic fibroma (AF) is a rare, benign odontogenic tumor of mixed epithelial and mesenchymal origin, accounting for approximately 2% of all odontogenic tumors. It commonly affects children and adolescents, with a strong predilection for the posterior mandible and typically presents within the first two decades of life. This report presents a case of a 3-year-old male child who was brought to the Department of Oral Medicine and Radiology by his parents with a chief complaint of a slow-growing, painless swelling in the mandible. Based on clinical, radiographic, and histopathological findings, a diagnosis of ameloblastic fibroma was established. The lesion was surgically enucleated and kept on regular follow up.

Keywords: Ameloblastic Fibroma, Epithelial, Ectomesenchymal

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1. Introduction

Ameloblastic fibroma (AF) is a rare, benign odontogenic tumor characterized by the proliferation of both epithelial and ectomesenchymal components without the formation of dental hard tissue. It was first described and named by Kruse in 1891 and later thoroughly defined by Thoma and Goldman in 1946.^{1,2} According to the World Health Organization (WHO) classification of odontogenic tumors, AF is categorized as a benign mixed odontogenic tumor, given its origin from both odontogenic epithelium and mesenchyme that resemble the enamel organ and dental papilla, respectively.³

AF accounts for approximately 1.5% to 2% of all odontogenic tumors and typically presents in the first two decades of life, with the posterior mandible being the most affected site.^{4,5} It often manifests as a slow-growing, asymptomatic swelling that may be discovered incidentally during routine radiographic examination or due to delayed eruption of teeth.

Histologically, the tumor comprises strands, cords, and islands of odontogenic epithelium embedded in a cellular mesenchymal stroma that mimics dental papilla.³ Importantly, there is no evidence of enamel or dentin formation, which

distinguishes AF from other mixed odontogenic tumors like ameloblastic fibro-odontoma or odontoma.⁴

Although AF is considered a benign lesion with a favourable prognosis, recurrence may occur, particularly if incomplete excision is performed.⁶ Moreover, in rare instances, malignant transformation into ameloblastic fibrosarcoma has been documented, underscoring the importance of long-term follow-up.^{6,7}

This case report highlights the clinical, radiographic, and histopathological features of ameloblastic fibroma in a 3-year-old male child, emphasizing the rarity of presentation at such a young age and the importance of early diagnosis and management in preventing complications and ensuring optimal facial growth and development.

2. A Case Report

A 3-year-old male child reported to the Department of Oral Medicine and Radiology accompanied by his parents with a chief complaint of swelling in the lower right back region of the jaw, persisting for the past three years. According to the mother, the swelling was first noticed as a small, soft,

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localized gingival growth in the posterior mandible. Over time, it got gradually increased in size, became bony hard and extended to the inferior border of the mandible which gave a visible extraoral bulge. The swelling remained painless, although gingival bleeding during mastication was reported. No history of trauma, systemic illness, or medication use was noted.



Figure 1: Extraoral photographs

On extraoral examination, **(Figure 1)** a single, unilateral, diffuse swelling was noted on the right posterior mandibular region, extended superoinferiorly from the right corner of the mouth to the inferior border of the mandible and anteroposteriorly from the midline to the body of the mandible. The swelling measured approximately 2 cm × 2 cm, was non-fluctuant, afebrile, and firm to bony hard in consistency.



Figure 2: Intraoral photograph



Figure 3: Intraoral photograph

Intraorally, **(Figure 2)** and **(Figure 3)** soft tissue examination revealed a well-defined, dome-shaped, reddish-pink to reddish-purple soft tissue mass, arising from the attached gingiva in the region of missing tooth 83, and extended over the crown of tooth 82, measuring approximately 1 cm × 1 cm. The labial surface of the mass appeared yellowish with erythematous margins, and it extended superoinferiorly from the alveolar ridge between teeth 82 and 84 to the labial vestibular region, and anteroposteriorly from the labial frenum to the distal aspect of tooth 84.

On palpation, the soft tissue growth was non-tender, soft, mobile, and pedunculated, whereas the underlying swelling was non-compressible, non-fluctuant, and firm to bony hard in consistency.

Hard tissue examination revealed the presence of teeth:

1. 55, 54, 53, 52, 51, 61, 62, 63, 64, 65 (maxilla) and 85, 84, 82, 81, 71, 72, 73, 74, 75 (mandible). Tooth 83 was clinically missing, and 84 showed caries.
2. Based on clinical presentation, a provisional diagnosis of Benign Odontogenic tumor was made. The differential diagnoses considered as Unicystic ameloblastoma, Ameloblastic Fibroma.

2.1. Investigations

2.1.2. Radiographic findings

An intraoral periapical radiograph (IOPA) **(Figure 4)** of the mandibular anterior region revealed fully visualised teeth 81, 82 and 84, with partial visualisation of 85. Developing crypts of permanent tooth buds 41, 42, and 43 were also noted. Tooth 83 was missing. A radiolucent lesion was seen in the missing tooth region with 83, and root caries was identified with respect to tooth 84.

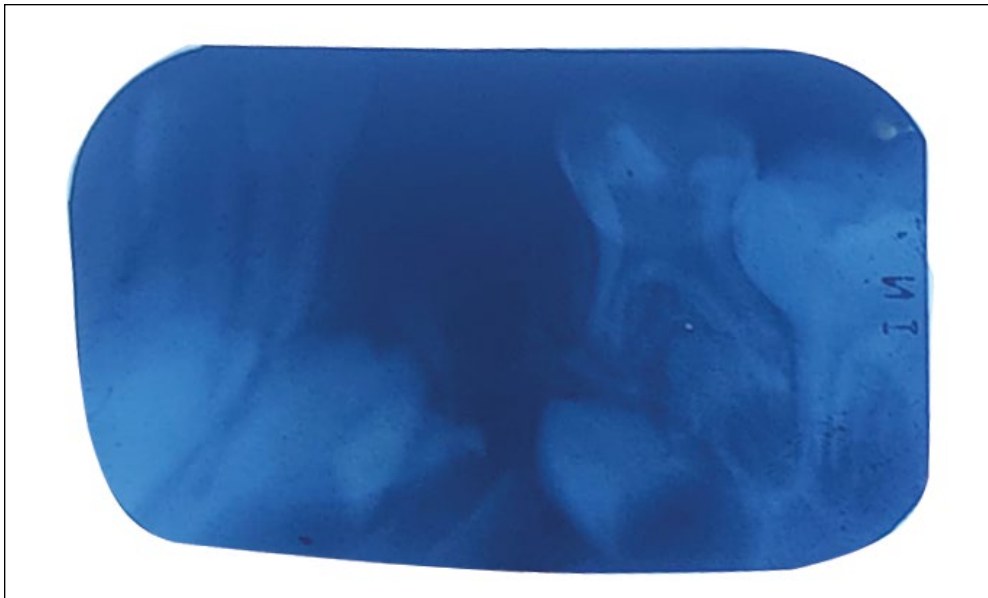


Figure 4: IOPA phototograph

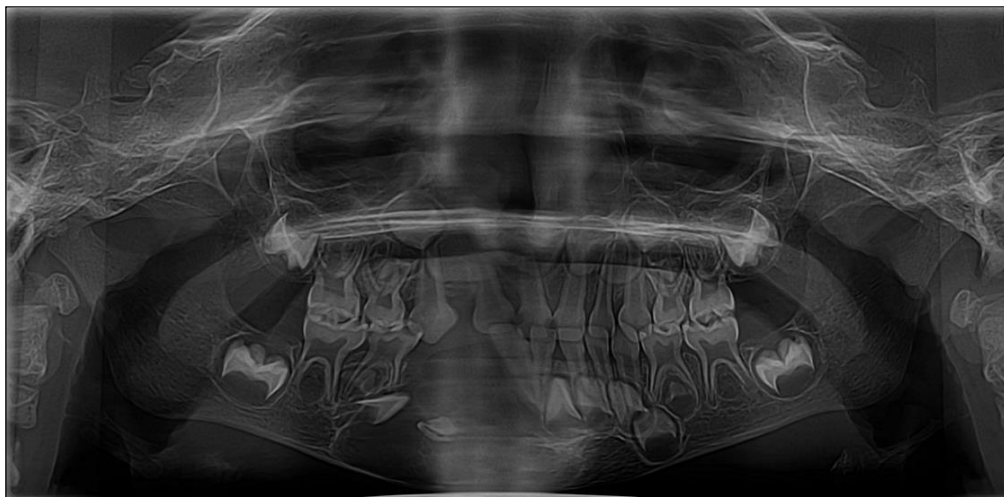


Figure 5: OPG phototograph

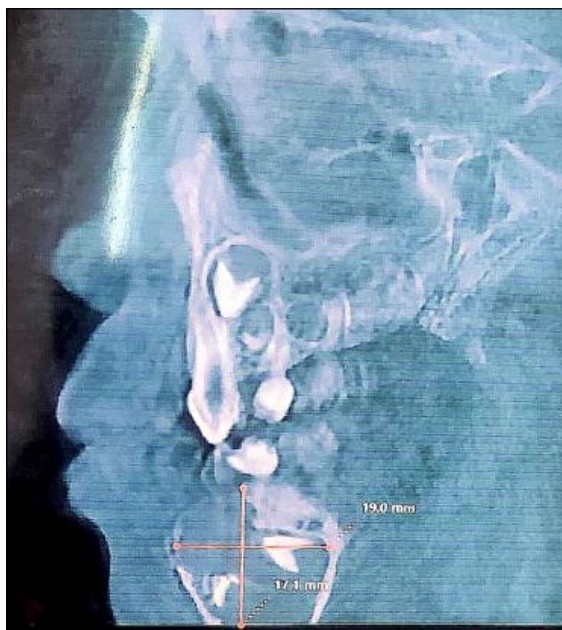


Figure 6: CBCT phototograph

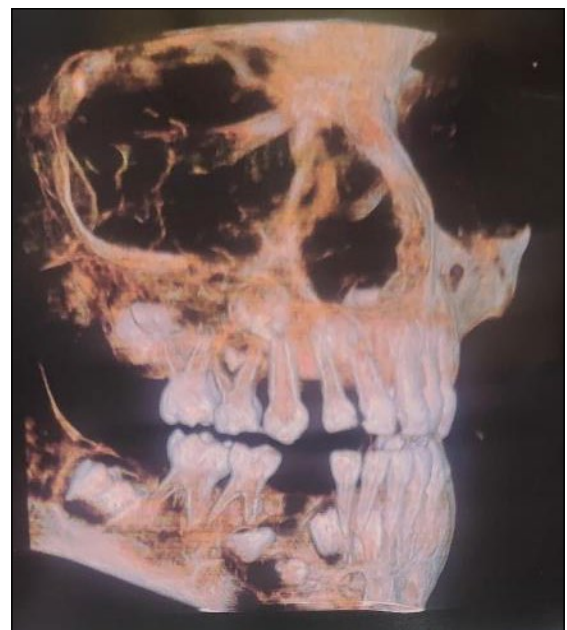


Figure 7: CBCT phototograph

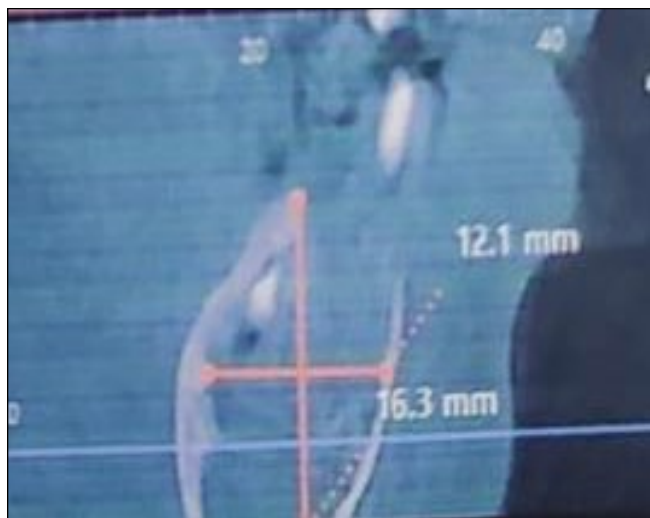


Figure 8: CBCT phototograph

To further evaluate the extent and internal architecture of the lesion, OPG (Figure 5) and Cone Beam Computed Tomography (CBCT) of the mandible was performed. (Figure 6), (Figure 7), (Figure 8) It revealed a well-defined, unilocular radiolucency in the right side of the mandibular region, measuring approximately 19mm (AP) $\times$ 13.4mm (ML) $\times$ 16.2 mm (SI). The lesion extended anteroposteriorly from the apical crypt of tooth bud 41 to the root apex of tooth 84, and superoinferiorly from the alveolar crest to the lower border of the mandible.

Internally, the lesion was completely radiolucent with a single, small, radiopaque tooth crypts of permanent teeth 42,43 were horizontally present. The lesion also caused displacement of the developing permanent canine (43) and thinning of the lower border of the mandible, along with buccal cortical plate expansion and thinning. The lingual cortical plate remained intact.

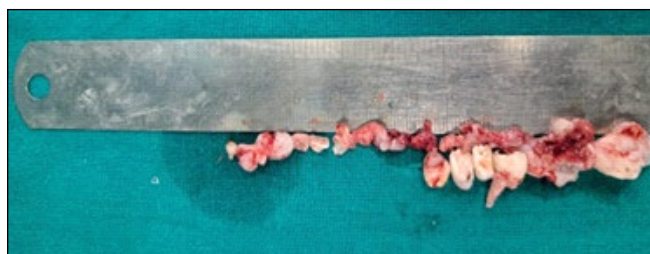


Figure 9: Excised specimen



Figure 10: Histopathological investigation



Figure 11: Post operative phototographs



Figure 12: Post operative phototographs



Figure 13: 1 Month follow up OPG

Based on radiographic features, Unilocular Ameloblastoma was given as radiographic diagnosis and differential diagnoses included Ameloblastic Fibroma. Incisional biopsy were

done which shows lining of “Ameloblastic Fibroma”. Patient was prepared for surgery and undergone extraction of 81,82,84,41,42,43 with marsupialization done. Excised specimen (**Figure 9**) was sent for histopathological investigation which confirmed the diagnosis of “Ameloblastic Fibroma”. (**Figure 10**) Thorough curettage with chemical cauterization done with Carnoy's solution and suturing done. (**Figure 11**), (**Figure 12**)

The patient was placed on a regular follow-up schedule, initially at one month postoperatively, followed by evaluations every six months. An OPG was taken at the one-month follow-up visit. (**Figure 13**) The one-month follow-up OPG shows signs of healing bone in the right posterior mandible following marsupialisation. The lesion has decreased in radiolucency and size, with signs of new bone formation and remodelling. No radiographic features suggest recurrence or aggressive behaviour at this stage.

2.1.3. Histopathological examination

1. The section shows both epithelial and mesenchymal elements. The epithelial component is arranged in the form of Narrow cords, strands, and follicle-like islands.
2. The islands resemble early dental lamina or enamel organ with peripheral columnar or cuboidal ameloblast-like cells and central loosely arranged stellate reticulum-like cells.
3. The mesenchymal stroma is Cell-rich, primitive, and resembles dental papilla-like connective tissue. Contains plump spindle-shaped cells in a myxoid background without significant mitotic activity.
4. No evidence of odontogenic hard tissue (dentin or enamel matrix) formation noted.
5. Overall features suggestive of “Ameloblastic Fibroma”.

3. Discussion

Ameloblastic fibroma (AF) is a rare benign odontogenic tumor of mixed epithelial and mesenchymal origin, comprising approximately 1.5% to 4.5% of all odontogenic tumors, as reported by Reichart and Philipsen in their comprehensive review of odontogenic tumors.^{8,9} It predominantly affects children and young adults, with most cases occurring in the first two decades of life, and the posterior mandible being the most common site.^{8,9} The current case is clinically significant due to its presentation in a 3-year-old child, which is far earlier than the commonly reported mean age of 14–15 years, emphasizing the need for early clinical suspicion and radiographic assessment in pediatric patients presenting with jaw swellings.

Radiographically, AF typically presents as a well-defined unilocular or multilocular radiolucency, frequently associated with unerupted or impacted teeth.^{9,10} This correlates with the present case, where a unilocular radiolucent lesion was observed displacing the developing permanent canine (43), with thinning and expansion of the buccal cortical plate but preservation of the lingual cortex. Such features

align with imaging descriptions from Shear and Speight, who highlighted the non-perforative yet expansile nature of AF, which can mimic other odontogenic cysts or tumors radiographically, including dentigerous cysts and unicystic ameloblastomas.^{10,11,12}

CBCT plays a pivotal role in delineating the extent of the lesion, its effect on adjacent structures, and surgical planning. In this case, CBCT revealed not only the lesion's proximity to multiple developing tooth buds but also cortical thinning without breach, aiding in the decision for conservative management. Literature supports the use of CBCT for assessing three-dimensional architecture, essential in pediatric cases where growth and facial symmetry are key considerations.^{12,13}

Histologically, AF is characterized by the presence of odontogenic epithelial islands, strands, and cords embedded within a cellular ectomesenchymal stroma that resembles the primitive dental papilla. The histological picture in our case confirmed these features, with stellate reticulum-like cells and ameloblast-like peripheral cells arranged in a collagen-rich stroma. These findings are consistent with the World Health Organization's 2022 classification of odontogenic tumors.^{3,13,14} Importantly, no dental hard tissue was observed, distinguishing AF from other mixed odontogenic tumors like ameloblastic fibro-odontoma or odontoma.^{15,16}

Interestingly, some cases of AF may exhibit areas of focal calcification or ossification, which blurs the histological distinction between AF and other lesions in the spectrum of odontogenic tumors. This supports the theory, proposed by Slootweg and Müller, that AF, ameloblastic fibro-odontoma, and odontoma may represent stages in a developmental continuum of odontogenic tumor progression.^{16,17} However, others argue that these are distinct entities rather than stages of the same lesion.

In terms of management, conservative surgical excision—through enucleation and curettage—is widely recommended, especially in young children, to minimize damage to adjacent structures and allow for continued jaw development. Our approach, including marsupialization and chemical cauterization with Carnoy's solution, aimed to reduce recurrence risk while preserving bone integrity. Literature suggests that Carnoy's solution effectively reduces residual tumor cells in the bone margins without compromising adjacent developing teeth if used judiciously.¹⁷

Despite its benign nature, AF has shown a notable recurrence rate ranging between 18% and 44%, particularly when treated conservatively.^{17,18} This has been demonstrated in several long-term follow-up studies, including by Zallen et al., who found that 33.3% of AF cases recurred, often several years postoperatively.^{17,18} Importantly, malignant transformation into ameloblastic fibrosarcoma (AFS) has been documented, especially in recurrent or long-standing cases. Zallen et al. reported that 11.4% of recurrent AF cases underwent malignant transformation, with one-third of AFS cases arising from pre-existing AF.^{17,18}

A 2020 systematic review by Upadhyaya et al. reinforced this concern, emphasizing that recurrent AF, particularly in older patients or those with delayed treatment, may carry a heightened risk of malignant change.¹⁸ Segmental resections have been suggested in high-risk or recurrent cases to reduce this risk, although such aggressive approaches must be balanced against functional and developmental considerations in pediatric patients.

Therefore, in this case, the decision to opt for conservative management was based on young patient age, lesion size, and absence of cortical perforation, but it was paired with strict long-term surveillance due to the established risk of recurrence and potential for malignant transformation. The one-month follow-up OPG already shows bone regeneration and lesion size reduction, suggesting a favourable early response, but periodic evaluation over several years is essential for definitive prognosis.

4. Conclusion

Ameloblastic fibroma is a rare benign odontogenic tumor that predominantly affects children and adolescents, with a predilection for the posterior mandible. This case underscores the significance of early diagnosis and management in a 3-year-old patient, an age much younger than the commonly reported onset. Prompt intervention through conservative surgical enucleation, marsupialisation, and chemical cauterisation with Carnoy's solution facilitated lesion regression and initial bone healing while preserving mandibular architecture.

However, despite its benign nature, ameloblastic fibroma carries a notable risk of recurrence and, in some cases, malignant transformation into ameloblastic fibrosarcoma, particularly in instances of incomplete removal or long-standing disease. This potential for aggressive behaviour reinforces the critical need for long-term surveillance, even in pediatric patients. Regular clinical and radiographic follow-up is essential for early detection of any recurrence or progression.

5. Source of Funding

None.

6. Conflict of Interest

None.

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