



A Rare Case of Ameloblastic Fibroma: Association of Central and Peripheral Clinical Forms

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How to cite this paper: Haitami, S., Fahim, O., Mahfoud, W., Sabir, E. and Ben Yahya, I. (2025) A Rare Case of Ameloblastic Fibroma: Association of Central and Peripheral Clinical Forms. *Open Access Library Journal*, 12: e13520.

<https://doi.org/10.4236/oalib.1113520>

Received: April 25, 2025

Accepted: June 13, 2025

Published: June 16, 2025

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Abstract

Background: An ameloblastic fibroma (AF) is a rare benign mixed odontogenic tumor, that commonly occurs during the first and the second decade of life, which is why it's often called a neoplasm of childhood and adolescence. It is composed of both epithelial and mesenchymal components, without any formation of calcified dental structure, and it can present in two clinical forms: a central (intraosseous) and a peripheral (extraosseous) variant. **Aim:** The aim of this paper is to report a rare case of AF that combined both clinical forms, in an adult patient, highlighting its diagnostic features and surgical management. **Case Presentation:** A 34-year-old patient presented to the Oral Surgery Department of the Dental Consultation and Treatment Center of Casablanca with a gingival overgrowth that had been present for two years, associated with a mild mandibular swelling in the premolar region. Histopathological examination of a biopsy confirmed the diagnosis of AF. Surgical treatment consisted of enucleation, curettage, and extraction of the involved teeth. **Conclusion:** This case illustrates a unique clinical presentation of AF involving both central and peripheral forms in an adult. It emphasizes the importance of thorough clinical, radiographic and histopathological evaluation for accurate diagnosis and appropriate surgical management of odontogenic tumors.

Subject Areas

Dentistry

Keywords

Ameloblastic Fibroma, Odontogenic Tumor, Benign Mixed Odontogenic Tumor

1. Introduction

Ameloblastic fibroma (AF), first described by Kruse in 1981, is a rare benign mixed odontogenic tumour [1] [2].

It accounts for approximately 2% of all odontogenic tumours [3]-[5]. It mostly affects the mandible, particularly the premolar-molar region, in about 80%, and it's associated in 75% of cases with impacted teeth [6]-[8].

AF occurs during the first and second decades of life, and has a slight male predilection, with a ratio of 1.4:1 [1] [3] [6] [8].

AF can present in two clinical forms: the central or intraosseous form, which arises within the jaw bones and represents the most common presentation, and the peripheral or the extraosseous, which occurs in the soft tissues, the latter being extremely rare, with very few cases documented in the literature [9].

We report an unusual and rare case of ameloblastic fibroma, presenting an association of both central and peripheral forms, in a 34-year-old patient, who belongs to an age group that is rare for this tumor. The patient was diagnosed and treated at the Department of Oral Surgery of the Dental Consultation and Treatment Center of Casablanca.

2. Case Report

A 34-year-old male patient, in apparently good general health, presented with a gingival hyperplasia that occurred two weeks after the extraction of a premolar, and that remains stable for two years. The patient also reported a mild mandibular swelling, for which he sought clinical examination in our department.

The extraoral examination revealed no pathological signs (**Figure 1**).



Figure 1. Pre-operative extra-oral image.

Intraoral examination showed a sessile inflammatory gingival growth, located at the site of the previously extracted tooth 45 (**Figure 2**). The lesion was firm, painless and non-bleeding upon palpation. A firm, painless mild vestibular swelling was also noted in the premolar region. Teeth 43 and 46 showed a negative pulp vitality test.



Figure 2. Intra-oral image showing a localized gingival budding.

The panoramic radiograph revealed a well-defined, multilocular radiolucent image, with a cyst like appearance, extending from the right retromolar region to tooth 33. A displacement of the 43's root and a narrowing with deviation of the inferior alveolar nerve were also observed (**Figure 3**).



Figure 3. Panoramic radiograph showing a well-defined multilocular radiolucent image.

Cone beam computed tomography showed a thinning of the buccal cortical plate and perforation at the crestal and the upper part of the vestibular plate of the mandibular ridge (**Figure 4**).

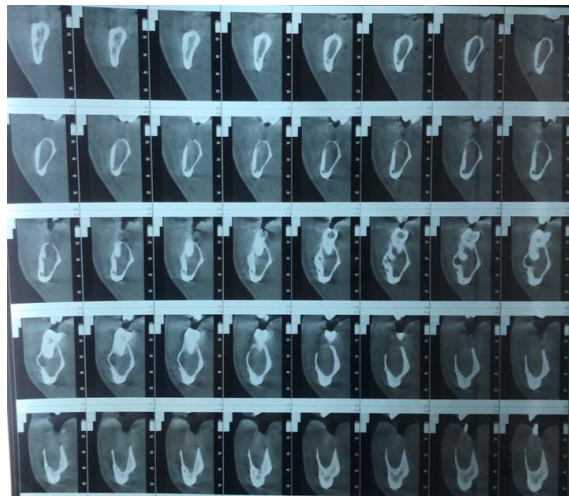


Figure 4. Pre-operative CBCT.

Differential diagnoses included: odontogenic keratocyst, ameloblastoma, radicular cyst, and a pyogenic granuloma for the peripheral component.

A biopsy for the gingival overgrowth was performed, which was consistent with an ameloblastic fibroma.

The treatment consisted of enucleation, curettage and extraction of teeth 46 and 43 under loco-regional anesthesia (**Figure 5**). Postoperative care included an amoxicillin-based antibiotic, corticosteroids, a step 2 analgesic and a chlorhexidine mouth-wash.

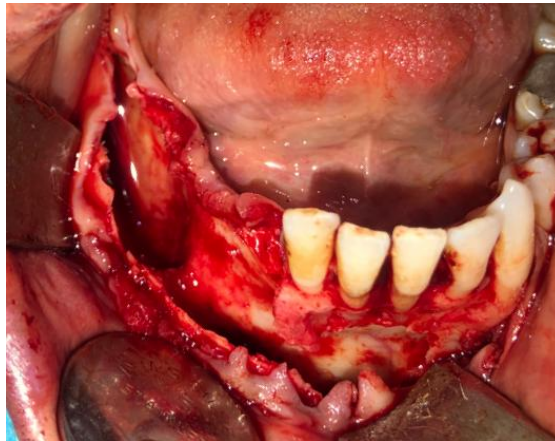


Figure 5. intra-operative image after complete enucleation and curettage.

The surgical specimen (**Figure 6**) was sent for histopathological examination. The sections showed a fibromatous ameloblastic tumor proliferation, with a fairly abundant connective-vascular stroma, exhibiting a fibro hyaline appearance in some areas. This proliferation consisted of small clusters of variable size, with a palisading arrangement at the periphery. The cells were monomorphic, without atypia or mitotic figures. This confirmed the diagnosis of ameloblastic fibroma.



Figure 6. Two surgical specimen, weighing a total of 20 grams and measuring $3 \times 2 \times 1$ cm and $2.5 \times 2 \times 1$ cm, and the extracted teeth.

At 8 month and 12 month radiological follow-up, the patient were asymptomatic, and showed mineralization and bone deposition in the cavity (**Figure 7**, **Figure 8**).



Figure 7. 8 month radiological follow-up showing the onset of bone regeneration.



Figure 8. 12 month radiological follow-up showing the bone regeneration of the cavity, with absence of pathological radiographic signs.

3. Discussion

AF is a rare benign mixed odontogenic tumor of childhood and adolescence, composed of a proliferating odontogenic epithelium within a mesenchymal tissue, resembling the dental papilla, without the formation of dental tissues such as enamel or dentin [1] [4] [10] [11].

It was first classified as a benign neoplasm, and known as soft odontoma in 1946 by Thoma and Goldman. In 1992, The world health organization (WHO) defined it as mixed neoplasm with varying degrees of dental tissue formation. In 2005, the WHO classified it under the category of mixed neoplasms as AF, with or without dental tissue formation (2,4). In 2017, the WHO clarified that hard tissue formation in these lesions, is usually the first stage of maturation corresponding to a developing odontoma. Consequently, It considered AF a true independent tumor, and

classified it under the category of odontogenic mixed tumors [12] [13], in which it remains listed according to the latest 2022 classification [14].

Two clinical forms of AF are observed: the central or intraosseous form and the peripheral form of the extraosseous one, which is very rare [9]. In fact, only 10 cases of peripheral AF have been described in the literature. Our case showed both forms, which may be considered as a central form with an extraosseous component involving the gingiva. The only similar case reported in the literature is a congenital ameloblastoma in a 2-week old infant [15].

AF may be asymptomatic in some cases [10], or it may present as a slow growing swelling with or without pain. The overlying mucosa can appear normal or present gingival budding or ulceration [6] [7]. These findings are consistent with those observed in our patient.

Radiographically, AF appears as a well-defined, unilocular or multilocular radiolucent lesion, with smooth and sclerotic borders [16]. It may also show cortical perforation, root displacement or resorption (1,8). As seen in our case.

The clinical differential diagnoses of the peripheral AF are represented by pyogenic granuloma, epulis, peripheral giant cell granuloma, focal fibrous hyperplasia, localised juvenile spongiotic gingival hyperplasia, and peripheral odontogenic fibroma [9]. As for the central variant, the differential diagnoses include ameloblastoma, central giant cell granuloma, odontogenic keratocyst, dentigerous cyst, odontogenic myxoma, and ameloblastic fibrosarcoma [8].

The positive diagnosis requires a histopathological examination [9].

The treatment of AF remains a subject of debate. It depends on the size of the lesion and whether it's primary or recurrent. It's usually treated with conservative approach (complete excision or enucleation), since it lacks aggressive and infiltrative behavior [7] [8]. The invasive approach such as marginal or segmental resection, is recommended when the tumor is recurrent and significantly large [16]. Since our patient is young, and had a primary AF, we opted for the conservative treatment.

AF has a good prognosis [12] [16]. The recurrence rate of AF ranges between 16.3% and 33.3%, depending on studies. It is often attributed to incomplete primary enucleation. AF presents a potential risk of malignant transformation into an ameloblastic fibrosarcoma (AFS) [9] [11]. A systematic review conducted in 2018 by Chrcanovic *et al.* reported a recurrence rate of 19.2% for central AF, 12.5% for the peripheral AF, and an incidence of 50% of secondary AFS from AF [5].

The average time between the treatment of an AF and the occurrence of an AFS is 55.1 months. This underscores the importance of long-term clinical and radiological follow-up [5]. At 12-month follow-up, our patient showed no signs of recurrence, and is still under observation.

4. Conclusions

We report a very rare case of AF, that has unusual characteristics, such as the age of onset, the presence of both clinical forms, and the involvement of the anterior

mandibular region

Regular long term follow-up, including both clinical and radiographic evaluation, is essential to detect any potential recurrence of malignant transformation.

From a clinical perspective, this case highlights the importance of considering AF in the differential diagnosis of mixed intraosseous and extraosseous lesions in adults, despite its rarity in this age group.

Further studies are needed to better understand the biological behavior and pathogenesis of AF, especially with dual clinical forms, to optimize its management.

Patient Consent

Both written and oral informed consent were obtained from the patient for the publication of this case report.

Conflicts of Interest

The authors declare no conflicts of interest.

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