Ameloblastic Fibro-odontoma - A Case Report of Two Uncommon Cases

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Abstract

Rationale: Ameloblastic fibro-odontoma (AFO) is a rare mixed odontogenic tumour with a preferential location in the posterior mandible and with a variety of radiological aspects. We report two clinical cases of AFO in two rare locations and with unusual radiological aspects. **Patient Concerns:** The first patient is a 37-year-old female with an asymptomatic intraoral swelling located in the anterior mandibular. The second patient is a 16-year-old girl with a chief complaint of missing maxillary canine. **Diagnosis:** Both cases were diagnosed with AFO. **Treatment:** For the first patient, a biopsy was performed, and for the second one, the lesion was surgically excised. **Outcomes:** The first patient is under regular surveillance and the other was followed up for one year without any evidence of recurrences. **Take-away Lessons:** Despite many efforts, the nature, histology and therapy of these lesions remain very confusing.

Keywords: Biopsy, cone-beam computed tomography, mixed tumour, neoplasms, odontogenic tumours

Introduction

Ameloblastic fibro-odontoma (AFO) is a benign mixed odontogenic neoplasm.[1] In the latest World Health Organization (WHO) Classification of Head And Neck Tumours, AFO is classified as a developing odontoma rather than a distinct tumour.[1]

AFO occurs predominantly in children and teenagers, with no predilection for gender. It is frequently associated with erupted or displaced teeth and can reach large sizes.[2] AFO has an equal predilection for the mandible or maxilla and favours the posterior areas.[3] Clinically, it presents as a painless swelling of the affected area, usually the posterior portion of the maxilla or mandible. Radiographs show a well-defined radiolucent area containing various amounts of radiopaque material of irregular size and form.[4]

In this article, we present two clinical cases of AFO in two rare locations, anterior mandibular and maxillary, and with unusual radiological aspects, in the first case, without an impacted tooth, and with a poorly limited polycyclic appearance, without calcification, and in the second case, the lesion looks like a developmental cyst in relation with an impacted canine, without calcifications.

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Case Reports

Case 1

A 37-year-old female reported to the Department of Oral Medicine and Oral Surgery at the University Dental Clinic of Monastir, Tunisia, with chief complaint of an asymptomatic intraoral swelling, approximately 3 cm at its widest point, located in the right anterior mandibular that had begun 4 months earlier with a progressive increase in size. The patient was asymptomatic, and she reported no pain or numbness. On palpation, the mass was asymptomatic and hard in consistency. Buccal cortical expansion was evident on palpation with displaced first and second mandibular incisors without any mucosal ulceration. The teeth in the affected area remain vital. No soft-tissue swellings or abnormalities were detected, and no grossly carious lesions were present [Figure 1].

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Dental history reported neither local trauma or infection at the lesion site nor medical history any remarkable systemic diseases.

Cone-beam computed tomography (CBCT) scan with coronal and 3-dimensional reconstruction was performed to further define the mass. The CBCT demonstrated a mixed, honeycomb-shaped, poorly defined bone, with destruction of the buccal cortex. The lesion extended from the mandibular central incisor to the mandibular canine. The lesion had displaced the mandibular central incisor and the mandibular lateral incisor [Figure 2].

Under local anaesthesia, a biopsy was performed using the punch biopsy technique [Figure 3].

Microscopically, the fragments are formed by islands of odontogenic epithelium surrounded by cellular connective tissue. Associated mineralised trabeculae have a dentin-like appearance. These histopathological findings led to the diagnosis of AFO [Figure 4]. Complete resection of the lesion is scheduled in 2 months.

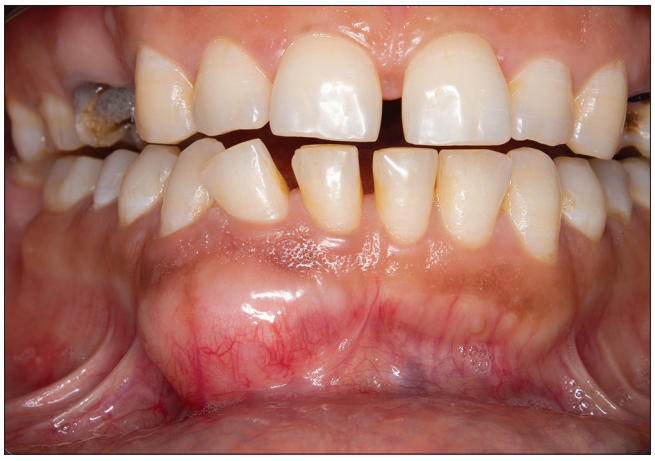

Figure 1: Intraoral mass extending from the mandibular central incisor to the mandibular canine

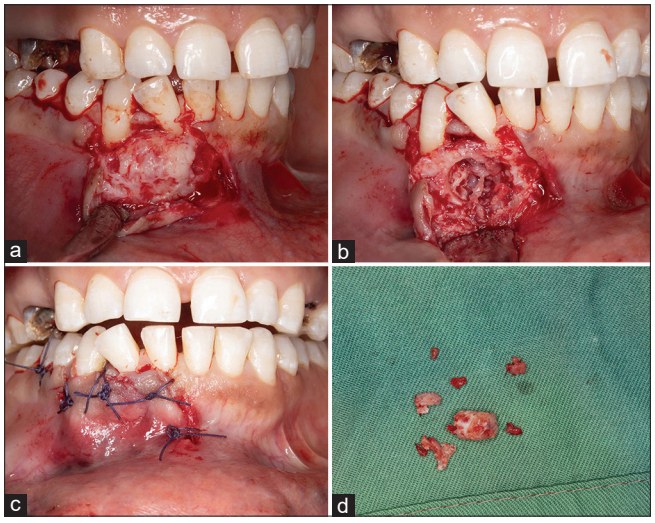

Figure 3: (a-c) Punch biopsy (d) Surgical specimen

Case 2

A 16-year-old girl reported to the Department of Oral Medicine and Oral Surgery at the University Dental Clinic of Monastir, Tunisia, with chief complaint of missing left maxillary canine. The patient was asymptomatic with the absence of intraoral or extraoral swelling. None of the teeth were mobile and the oral mucosa appeared normal and showed no signs of ulceration [Figure 5a]. CBCT scan was performed and showed a homogeneous, radiolucent, well-defined lesion measuring approximately 1 cm inserted into the neck of the impacted maxillary canine [Figure 5b and c]. The lesion was surgically excised under local anaesthesia with preservation of the impacted canine [Figure 6]. The excised tissue was subjected to histopathological examination.

Microscopically, the tissue specimen showed loose connective tissue with a myxoid base and few cells. In some areas, this tissue contains well-circumscribed islands of epithelial cells, some of which are calcified. These histological findings were suggestive of AFO [Figure 7]. During the follow-up of one year, the wound had healed well, and no recurrence of the tumour was observed clinically and radiographically.

Discussion

AFO is a relatively rare, benign odontogenic tumour whose etiopathogenesis is controversial, with limited cases documented

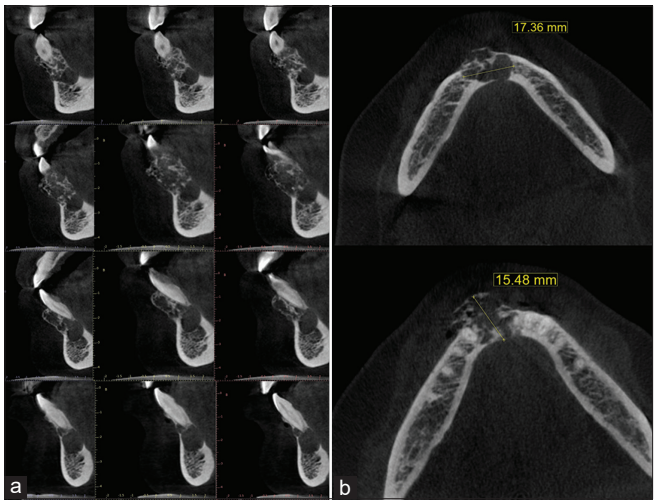

Figure 2: (a) Preoperative cone-beam computed tomography scan coronal view of a mixed lesion. (b) Three-dimensional reconstruction revealing a mixed lesion extended from the mandibular central incisor to the mandibular canine

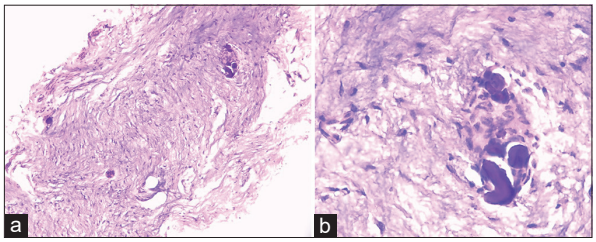

Figure 4: Histopathological specimen reveals the presence of myxoid cell-rich stroma resembling the dental papilla containing islands of odontogenic epithelium with calcification (a: H&E ×40, b: H&E ×400)

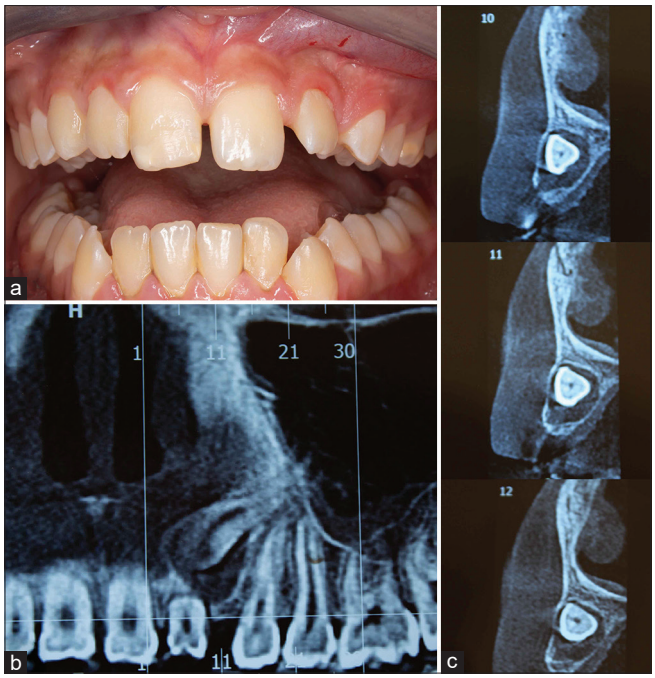

Figure 5: (a) Intraoral view showing the absence of the left maxillary canine, (b and c) Preoperative cone-beam computed tomography scan showing a radiolucent lesion inserted into the neck of the impacted maxillary canine

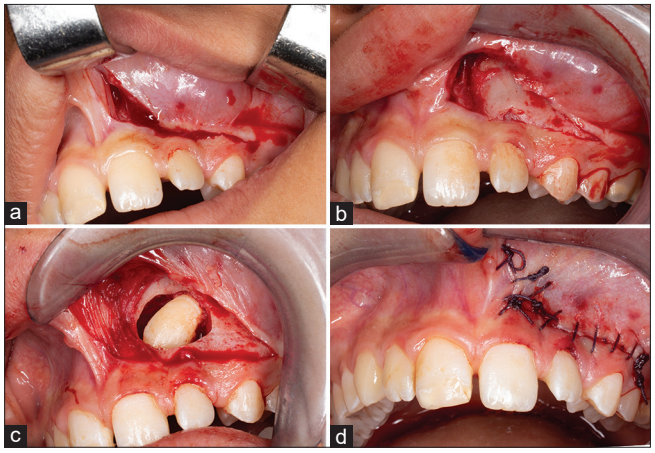

Figure 6: (a) Arciform incision, (b) Full-thickness flap, (c) Lesion removal, (d) Simple continuous sutures

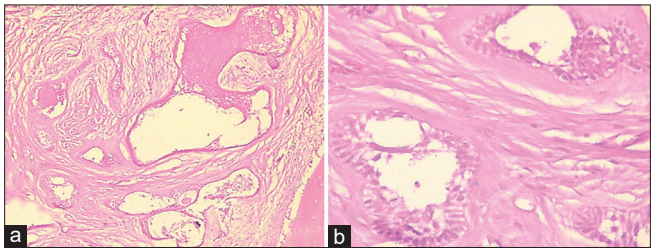

Figure 7: Photomicrograph of a section of the specimen showing islands of odontogenic epithelium showing peripheral palisading embedded in myxoid cell-rich stroma showing dentin (in the right side) (a: H&E × 200, b: H&E × 400)

in the literature.[2] The frequency of AFO ranges from 1% to 3%, considering all odontogenic tumours.[2] There is no gender predilection, with the lesion being equally found in the mandible and maxilla, normally in the molar region.[4] It affects young patients, especially in the first two decades, with a mean age of 11.5 years.[4] In our first case, the age of the patient was 37 which adds peculiarity to our clinical situation. It is generally considered a slow-growing central jaw tumour; occasionally, the tumour exhibits marked swelling, which results in facial disfigurement. In a systematic review published by Atarbashi-Moghadam *et al*., in 2019,[5] 11 articles (reporting 14 cases) were selected. Patients' mean age was 13.75 years (male/female = 1.8). The swelling was reported in 78.57% of the cases, pain in 28.57%, but 21.42% were asymptomatic.[5]

An update of a systematic review, published by Chrcanovic and Gomez in 2017,[6] was carried out by our team including, to the best of our knowledge, all cases of AFO published from August 2016 to December 2023, with a focus on localisation and radiological appearance, both of which give a rarity to our clinical cases. A total of 229 patients presented with AFO, 57.2% of whom were men. The mandible was more affected than the maxilla, with 65.5% of cases; in our update, of the 18 cases added, 15 were in the mandible with only one case in the anterior region [Table 1].

Concerning the radiological aspect of the AFO, which was unusual for the two clinical cases, and referring to Table 1, indeed 70.3% of AFOs cited in the literature present centro-lesional radio opacities, which is not the case for our patients, and especially for the second case where we observed a radiolucent image associated with an impacted canine without calcification giving the appearance of a developmental cyst. The majority of cases presented unilocular lesions (70.3%), associated in 59.4% of cases with impacted teeth and causing displacement or unerupted tooth, in 78.2% of cases. These radiological characteristics are not valid for our first clinical case, where the lesion was multilocular, with a mottled appearance of bone, not associated with impacted teeth or caused tooth displacement, orienting the diagnosis in favour of osteolytic giant cell lesions such as central giant cell granuloma, giant cell tumour or brown tumour or even for malignant lesions based on the mottled appearance of the bone and the lack of defined lesion limits. Nevertheless, confirmatory diagnosis is made according to a microscopic study, demonstrating islands of odontogenic epithelium embedded in cell-rich ectomesenchyme similar to dental papilla [Table 1].

Concerning the classification of AFO, more than 4 years have passed since the last WHO classification of head-and-neck tumours, some complex issues remain unclear. A study published by Soluk-Tekkesin and Vered in 2021.[7] A total of 23 well-documented AFOs were analysed and they suggested to reconsider at least a part of the AFOs, especially those in patients younger than 13.5 years with lesions of 2.1 cm and larger in diameter, as representing true tumours rather than

Table 1: Demographic and clinical features of ameloblastic fibro-odontomas described in the literature

	n (%)
Sample size (*n*) age (year)	229 (1–55)
Gender	
Men	131 (57.2)
Women	94 (41)
Unknown	4
Associated with tooth	
Yes	136 (59.4)
No	34 (14.8)
Unknown	59
Jaw	
Maxilla	73 (31.9)
Mandible	150 (65.5)
Unknown	7
Cortical bone perforation	
Yes	42 (18.3)
No	112 (48.9)
Unknown	74
Tooth displacement/unerupted	
Yes	179 (78.1)
No	21 (9.2)
Unknown	29
Locularity	
Unilocular	161 (70.3)
Multilocular	26 (11.3)
Unknown	42
Radiopacities	
Yes	161 (70.3)
No	25 (10.9)
Unknown	43
Treatment	
Excision/curettage	4 (1.7)
Enucleation	179 (78.1)
Marginal resection	6 (2.6)
Segmental resection	7 (3)
Unknown	9

developing odontomas. For AFO in youngsters, conservative therapy is recommended due to the lesion's benign nature and low recurrence incidence. When a tooth is associated with a lesion, it may or may not be extracted, depending on the feasibility of keeping it in place and ensuring that no neoplastic leftovers remain after the disease is removed completely.[8]

Conclusion

Given the non-pathognomonic clinical aspects of AFO and the various radiological aspects, the positive diagnosis is mainly based on anatomopathological examination of the surgical specimen or biopsy of the lesion if it is too extensive while supporting the results of histological examination with clinical and radiological data.

Declaration of patient consent

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

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Nil.

Conflicts of interest

There are no conflicts of interest.

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