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Case Report

# Ameloblastic Fibro-odontoma with a Predominant Radiopaque Component

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## Abstract

Ameloblastic fibro-odontoma (AFO) is a rare odontogenic tumor. Initially believed to be a lesion similar to ameloblastic fibroma (AF), it is now considered as a separate entity in the WHO odontogenic tumor classification. Commonly associated with a painless swelling and an associated absence of eruption of a tooth, AFO presents as a mixed radiopaque and radiolucent lesion in the younger population with a predilection for the posterior region. Histologically, it shows the characteristics of an immature complex odontoma with irregularly arranged enamel, dentinoid, cementoid-like structures, and ectomesenchymal tissue. The following case report describes a case of AFO with a predominantly radiopaque component and briefly discusses the available literature pertaining to this rare entity.

**Keywords:** Ameloblastic fibro-odontoma, ameloblastic fibroma, radiopaque

## Introduction

Odontogenic tumors are a pathology occasionally encountered by the practicing clinician. Studies from North America seem to indicate that odontogenic tumors represent approximately 1% of all accessions in oral pathology laboratories.[1] While ameloblastomas and odontomas are encountered more frequently, ameloblastic fibro-odontoma (AFO) is rare, accounting for 1%–3% of all the odontogenic tumors.[2] AFO was defined as a lesion similar to ameloblastic fibroma (AF), but also showing inductive changes that lead to the formation of both dentine and enamel.[3] However, AFO is presently defined by the World Health Organization as a neoplasm consisting of odontogenic ectomesenchyme resembling the dental papilla, epithelial strands, and nest resembling dental lamina and enamel organ in conjunction with the presence of dentine and enamel.[4]

The pathogenesis of AFO appears to be from an abnormal proliferation of odontogenic epithelium from a permanent tooth germ, which exerts an organizing effect on the mesodermal element with the formation of calcified dental tissues.[5]

The following is a case report of a male patient with a large AFO lesion in the posterior mandible with a predominant radiopaque component.

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## Case Report

A 19-year-old male patient presented to us with a chief complaint of a painless swelling in the lower right side of the mouth since 6 months. Examination revealed a small hard swelling in the right mandibular buccal and lingual sulcus and absence of the tooth 47. History revealed that the patient had not undergone any extraction for the tooth in the past and that the tooth had not erupted in the mouth. Opposite arch 37 was erupted and the 38 was found to be impacted clinically. The overlying mucosa was normal, the nerve function was well preserved, and the tooth 46, adjacent to the swelling exhibited no pathologic mobility or pockets. There was no draining sinus or fistula formation. Aspiration was performed to rule out any vascular malformation and on aspirate being negative; an incisional biopsy was performed. An incisional biopsy report of odontomes was received, and a cone-beam computed tomography (CBCT)

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was advised. CBCT revealed the presence of a large well-defined radiolucency extending from the mesial root of 46 to the inferior aspect of the ascending border of the ramus. Measuring approximately 6 cm × 3 cm in the greatest dimension anteroposteriorly and superoinferiorly, it could be seen extending till the inferior border of the mandible. Teeth 47 and 48 were involved within the radiolucency, and the inferior alveolar canal was deflected inferiorly. Correlating clinically, a mild buccolingual cortical expansion could be appreciated. The patient gave no neurological disturbances secondary to inferior alveolar nerve compression. The bulk of the radiolucency was interspersed with well-defined areas of radiopacity [Figure 1a and b].

A surgical excision of the lesion was planned owing to the well-defined nature of the radiolucency and an incisional biopsy report of an odontome. Informed consent was obtained from the patient, and the surgical procedure was performed under general anesthesia with oral intubation. Due to the proximity of the lesion to the inferior border of the mandible and an associated risk of mandibular fracture, an extraoral submandibular incision was given. Surgical exposure of the lesion revealed an expanded buccal cortex which was not breached [Figure 2a]. A bony window was created in the buccal cortex, and the lesion was excised in a piecemeal fashion owing to the large dimensions of the lesion [Figure 2b]. 46 was extracted intraoperatively based on the severe bone loss in relation to its distal aspect. The resulting surgical defect was evaluated [Figure 2c]. Owing to the thin margin of the inferior border of the mandible which was left behind, a risk for pathologic fracture was suspected. A 2.5 mm titanium reconstruction plate was hence placed along the body and the ramus of the mandible to support it [Figure 2d]. The closure was done in layers, and the specimen was sent for histopathologic examination. The postoperative recovery of the patient was uneventful [Figure 3a-c].

### Histologic examination

The specimen consisted of multiple hard tissue bits, the largest hard tissue bit measuring around 10 mm × 5 mm × 3 mm and 8 mm × 5 mm × 3 mm. The H and E stained soft tissue sections exhibited connective tissue consisting of round to ovoid-shaped odontogenic islands which were peripherally lined by tall columnar cells arranged in a palisading pattern with reversal of polarity and subnuclear vacuolization. The center of the odontogenic islands showed the presence of stellate reticulum like cells. The islands were surrounded by a clear zone which was previously occupied by the enamel matrix. Areas of hyalinization resembling dysplastic dentin could also be seen [Figure 3d and e].

## Discussion

First described by Hooker in 1967,[5] AFO has since been extensively studied in literature. However, due to its rare occurrence, the available literature has remained restricted to mainly single case reports. Some investigators have hypothesized that AF, ameloblastic fibrodentinoma (AFD), and AFO could represent a single entity in different stages of development: AF or AFD would evolve to AFO as they mature and the latter could differentiate into odontoma. However, when clinical characteristics of these lesions are analyzed, AFO is more frequent in a younger age group with a mean age of 9.6 years, than AF, which affects individuals with a mean age of 14.8 years, thereby in disagreement to the continuous differentiation hypothesis.[2] Philipsen *et al.* suggested the hypothesis of two lines of development for AF, AFD, and AFO. A hamartomatous line was represented by AFO as a primary stage of odontoma whereas a neoplastic line comprised AF and AFD.[5]

The diagnostic difficulties for AFO can be due to its resemblance to calcifying epithelial odontogenic tumor, adenomatoid odontogenic tumor, AFD, and AF. The differential diagnosis between AFO and AF is made based on the presence or absence of elements indicative of tooth germ differentiation (enamel or dentin). While AFO exhibits evidence of tooth germ differentiation, AF does not.[6] However, differentiating AFO from AFD can be difficult as both present as well-circumscribed, expansile radiolucencies on radiographs that generally contain a few solitary or multiple small radiopaque foci with irregular sizes that form a calcified product in the lesion and are often associated with impacted tooth/teeth. Consequently, a histopathological diagnosis is typically required to distinguish AFD from AFO.[7]

These tumors are commonly seen in the younger population with 98.8% cases occurring before the age of 20 years. While certain authors claim no gender preference,[6] a recent systematic review by Chrcanovic and Gomez[8] found that AFO was more prevalent in men than women (1.4:1). The lack of any clinical complaints such as pain, pus discharge, or significant swelling could be a possible reason for their delay in diagnosis or an incidental diagnosis during a routine dental check-up.

Radiographic appearance of AFO may be in the form of a well-defined radiolucent area containing variable amounts of radiopaque materials of irregular size and form. About 86% lesions are found to be associated with impacted teeth. Root resorption of the adjacent teeth may also occur, the majority of the teeth being deciduous teeth.[8] The presence of a predominant radiopacity within the radiolucency was consistent with our case.

Despite the considerable size the lesions of AFO may attain, pain and/or paresthesia are unusual symptoms, although tooth displacement may occasionally occur.[2] Our patient presented with no complaints of pain or parsesthesia despite evidence of downward displacement of the inferior alveolar canal due to the lesion. In addition, our patient also did not exhibit any tooth displacement in relation to 46.

Araki *et al*.[9] studied the diverse calcification patterns seen in radiographs of patients with AFO. He categorized the calcification patterns based on their appearance and their

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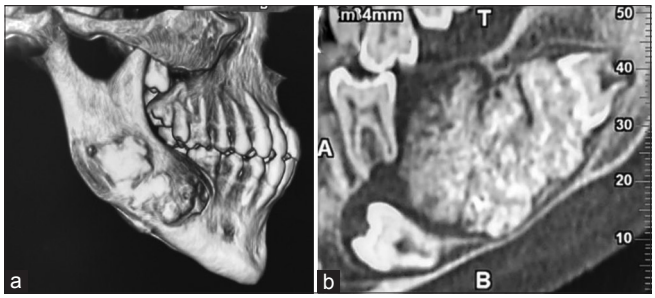


**Figure 1:** (a) Three-dimensional reconstruction showing the extent of the tumor. (b) Sagittal computed tomography section exhibiting impacted 47, 48 within the mixed lesion

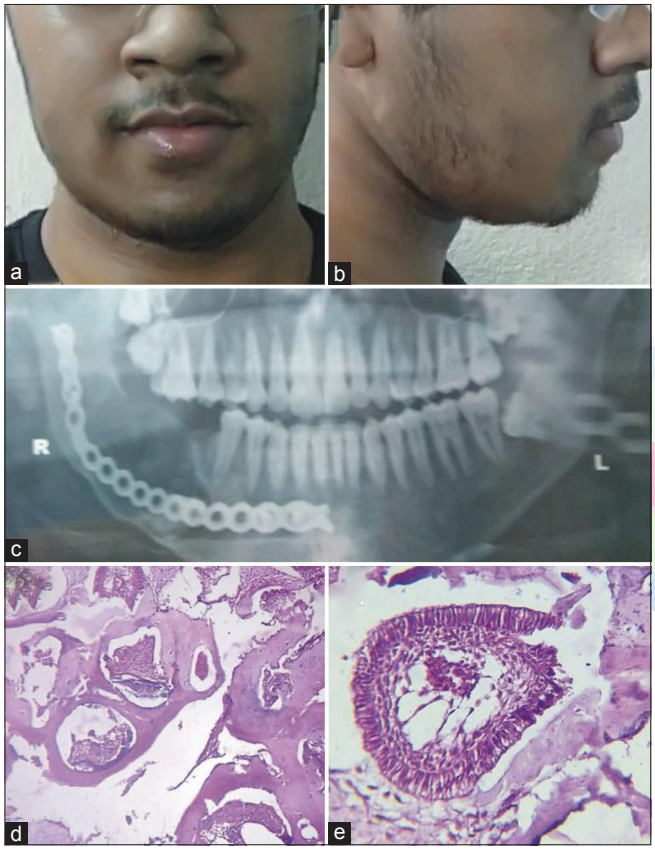


**Figure 3:** (a and b) 6th month postoperative frontal and profile photographs. (c) 2nd month post-operative orthopantomogram with the residual defect and the reconstruction plate in place. (d and e) Histological features at ×10 and ×40 magnification

location within the radiolucency. While the appearances were categorized into lucent, sand, cluster, blended and tooth-like, the location was categorized as peripheral, central, or full. The radiographic features of our patient could be hence categorized as cluster appearance located centrally within the lesion. However, the fill of the lesion was found to be extensive with a minimal radiolucent rim.

A conservative surgical approach in the form of excision or enucleation has been found to be satisfactory for these well-encapsulated lesions. Sporadic recurrences of AFO have been attributed to the inadequate surgical removal at the time of initial treatment. The use of carnoy's solution and cryosurgery has also been reported to reduce the chances for recurrence.[2]

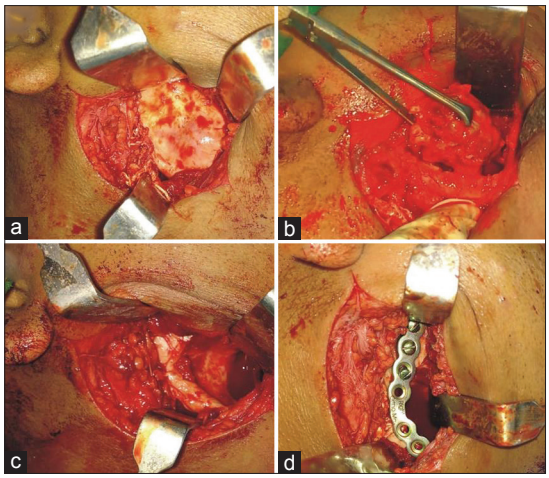


**Figure 2:** (a) Surgical exposure of the lesion. (b) Tumor excision. (c) Defect post excision. (d) Reconstruction plate fixation

Large lesions pose a risk for pathologic fracture during surgical removal because of thinning of the bone. The lesion should hence be removed in small pieces by sectioning the mineralized tissue to preserve the bone and minimize the amount of bone lost. Due to the large size of the lesion in this case, a large surgical defect was produced with a resultant increased risk of pathologic fracture. To support the mandibular bone in this region, a 2.5 mm reconstruction plate was adapted and plated along the mandible extending from the ramus to the body.

Another clinical decision pertaining to enucleation of AFO lies in the fate of the teeth around the lesion. Zouhary *et al*.[10] advocated that if the teeth do not interfere with the enucleation of the tumor, there is no reason to remove them, with the possibility of a spontaneous eruption occurring later. In case of our patient, the impacted 47 and 48 were completely embedded within the lesion and were hence removed along with the tumor.

A recent systematic review by Chrcanovic and Gomez revealed that unilocular lesions appeared to recur more frequently compared to multilocular varieties of AFO. Furthermore, multilocular lesions were found to be a more common feature of AFD compared to AFO.[8]

AFO is a rare group of odontogenic tumors with a good prognosis. The involved teeth can be left behind to allow spontaneous eruption into the oral cavity as long as it does not interfere or compromise with the complete excision of the lesion. The well-defined nature of this lesion and meticulous surgery makes excision easy and minimizes chances for recurrences further.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other

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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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## Conflicts of interest

There are no conflicts of interest.

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