

Ameloblastic Fibro-Odontoma of the Maxilla in 4th Decade: An Infrequent Case Presentation

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Abstract

Ameloblastic fibro-odontoma represents a hamartomatous rather than a neoplastic odontogenic lesion. It is a mixed odontogenic lesion with the histologic features of both ameloblastic fibroma and complex odontoma. This report depicts the case of a 45-year-old man with Ameloblastic fibro-odontoma on the right anterior maxillary front tooth region. On histological examination, sections of excisional biopsy revealed small islands of odontogenic epithelium with peripheral tall columnar ameloblast-like cells with reversal of polarity, central stellate reticulum like cells, and conglomerate mass of enamel and dentin arranged in a disorganized pattern seen in close proximity of the epithelial islands. These features led to the diagnosis of Ameloblastic fibro-odontoma.

Keywords: *Ameloblastic Fibro-Odontoma; Odontoma; Mixed Odontogenic Lesion; Hamartomatous; Neoplasm*

Introduction

Ameloblastic fibro odontoma (AFO) represents a hamartomatous rather than a neoplastic odontogenic lesion. There are well-documented examples, however, of this tumor exhibiting progressive growth and causing considerable deformity and bone destruction. Such lesions appear to be true neoplasms [1].

The AFO is a rare benign odontogenic lesion defined as a tumor with the general features of ameloblastic fibroma but that also contains enamel and dentin [3].

The AFO with an incidence of 1 to 3% [4] is rare in adults usually seen in children with an average age of 10 years. They are found to occur in the posterior region of the jaws with no significant sex predilection. They are usually asymptomatic and can be diagnosed in a radiograph taken to check when a tooth has failed to erupt [1].

The lesions present as well-circumscribed, unilocular or multilocular radiolucent lesions with radiopaque border at their periphery, radiographically. They may contain a variable amount of calcified material with the radiodensity of tooth structure. The calcified material within the lesion may appear as multiple, small radiopacities or as a solid conglomerate mass [1]. Most cases are associated with an unerupted tooth located in the periphery [17]. Treatment of AFO includes conservative surgery with enucleation [5]. Prognosis is excellent [6].

Case Report

A 45-year-old male patient reported to the Department of Oral and Maxillofacial Surgery, King Georges' Medical University, Lucknow with the chief complaint of swelling in the upper front tooth region for 20 years.

History of present illness revealed a swelling which was present for 20 years. On extraoral examination, a diffuse swelling was seen on the right cheek region adjacent to the ala of the nose. Intraorally, a bony hard swelling was seen in the maxillary anterior tooth region involving the cuspid and the first bicuspid. It was approximately, 2 cm x 1 cm in size, pink in color and oval in shape. It was non tender and no discharge of any kind was noticed. Radiographically, a well-defined mixed radiolucency was seen near the roots of cuspid and the first bicuspid. Based on the clinic-radiographical findings, a provisional diagnosis of Ossifying fibroma and Osteoma was made.



Figure 1: The orthopantomogram showing a mixed radiolucency in the anterior maxillary region.

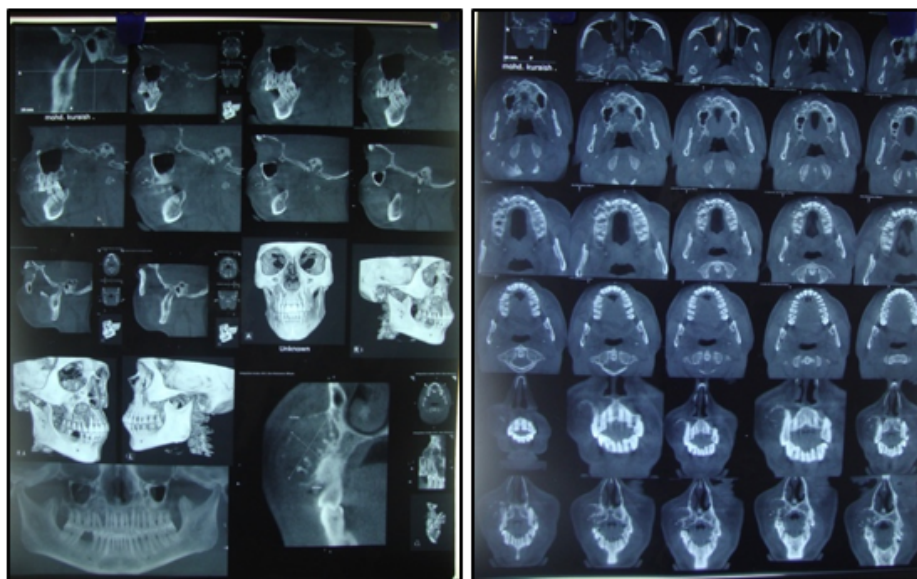


Figure 2

The lesion was surgically excised and sent for histopathological investigation. Surgical specimen was fixed in 10% formalin and subjected to pathological analysis. Gross examination revealed a hard tissue specimen with two teeth attached measuring 2 x 4.5 x 2.5 cm in size, irregular nodular shape and surface with ischemic areas, rough in contour, grayish brown in color and hard in consistency. The teeth were removed, specimen sectioned into two halves and kept for decalcification.



Figure 2: Grossing specimen revealing the presence of the lesional tissue in toto along with two attached teeth specimen.

The hematoxylin and eosin stained sections revealed narrow cords and small islands of odontogenic epithelium in a loose connective tissue stroma. The epithelial islands show peripheral tall columnar ameloblast-like cells with reversal of polarity and central stellate reticulum like cells. Few islands show squamous metaplasia along with cystic degeneration. Conglomerate mass of enamel and dentin arranged in a disorganized pattern is also seen adjacent to the ameloblastic epithelium.

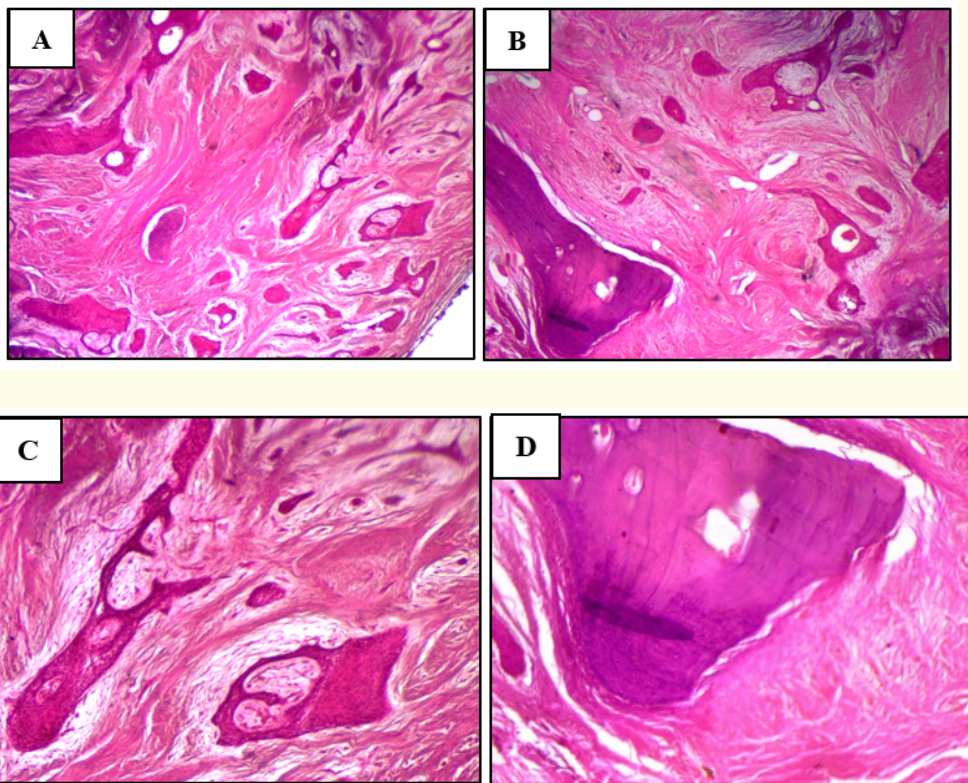


Figure 3: Photomicrographs of Ameloblastic fibro-odontoma.

- A: Photomicrograph of section showing strands, cords and nests of odontogenic epithelium in form of isolated islands in a moderately cellular fibrous connective tissue stroma (H & E stain, 4x).
- B: Photomicrograph of section showing intermediate zone between the soft tissue component and the hard tissue composed of dentin and enamel (H & E stain, 4x).
- C: Photomicrograph showing numerous epithelial islands with peripheral tall columnar cells showing reverse polarity of nucleus simulating ameloblasts and central loosely arranged cells resembling stellate reticulum (H & E, 10x).
- D: Photomicrograph of higher magnification of the hard tissue component composed of dentin and enamel spaces (H & E stain, 10x).

Discussion

Ameloblastic fibroodontoma has typically been classified as a benign mixed odontogenic tumor. Very few cases have been reported in the literature so far. 0.3% to 1.7% of odontogenic tumors are known to be AFO [7]. 4.6% of the total cases reported were pediatric patients [7]. There is no sex predilection. Posterior border of the mandible (2.4: 1) is more frequently involved [8]. The present case involved the anterior maxillary region which is a rare finding.

The AFO has been confused with the odontoma, although a clear distinction between the two was finally drawn by Hooker in 1967 at the annual meeting of the American Academy of Oral Pathology [2]. Slootweg also cleared the confusion with another entity called ameloblastic fibroma and concluded that the ameloblastic fibroma is a true neoplasm and does not differentiate into an AFO [2]. Slootweg and Takeda also concluded that AFO should not be considered as hamartomatous as there are cases of AFO showing true neoplastic behavior and the existence of malignant variants [9,10].

Buchner, *et al.* [11] in 2013 reported 114 cases, showing a mean age of 9.6, with a male-to-female ratio of 1.85:1. Posterior mandible was found to be the most common site. They witnessed a mixed radiolucent pattern in most of the cases [11]. A similar study was done by Kirjavainen, *et al.* [12] in the year 2016. A total of 108 cases were reported showing a mean age of 6.3 in females and 9.6 in males, male-to-female ratio of 1:1.62. Posterior mandible was again found to be most commonly affected [12]. An updated systematic review of 215 cases stated in the literature was published by Chrcanovic BR, *et al.* [13] in the year 2017. They reported an average mean of 9 ± 5.4 . Only 4 lesions were reported to have an age predilection of over 30 years. A male to female ratio of 1.3:1 was seen. Mandible was more commonly involved than the maxilla. A mixed unilocular radiolucency was seen [13]. The case reported in our institution showed gender and radiographic concomitance with the above-mentioned literature. It was seen in a 45 old male patient present in the anterior maxillary region making it an exceptional case.

CT scan findings shows a nearly well-defined radiolucency with radiopacity is present on right side of maxilla extending from mesial to canine to distal to second premolar. It shows loss of buccal cortical plate suggestive of an odontogenic tumor. Maxillary sinus is within anatomical limits. The findings are suggestive of Adenomatoid odontogenic tumor.

It is very easy to diagnose an ameloblastic fibroma-odontoma when it has a usual presentation of age, location and radiological pattern. Our case presented atypically but was diagnosed on the basis of histological presentation of small islands of odontogenic epithelium with peripheral tall columnar ameloblast-like cells with reversal of polarity, central stellate reticulum like cells, and conglomerate mass of enamel and dentin arranged in a disorganized pattern seen in close proximity of the epithelial islands [1].

AFO should be differentiated from complex and compound odontoma, ameloblastic fibroma, ameloblastic fibro dentinoma, odontoameloblastoma, calcifying epithelial odontogenic tumor, adenomatoid odontogenic tumor, and calcifying odontogenic cysts to conclude to a diagnosis [14]. If there is dentin formation, the lesion should be diagnosed as (AFD), whereas a lesion similar to ameloblastic fibroma showing inductive changes that lead to the formation of dentin and enamel both is diagnosed as AFO [15].

A conservative surgical approach should be the treatment of choice for Ameloblastic fibro-odontoma. Sporadic recurrences of AFO have been attributed to the inadequate surgical removal at the time of initial treatment [16]. There is a controversy in the literature regarding extraction or retaining the associated tooth bud in the case of AFO [16]. It has been stated that associated tooth bud excised with the lesion is important to avoid recurrence.

Sl. No.	Name	Striking Features	Differentiating Features
1.	Complex odontoma	Mass of disorganized tissue seen	Presence of ameloblastic islands
2.	Compound odontoma	Denticles and tooth resembling calcifications seen	Calcifications never resembles a tooth and ameloblastic islands present
4.	Ameloblastic Fibroma	Islands and strands of epithelial cells in a loose connective tissue stroma resembling primitive dental papilla	Calcifications seen and myxoid connective tissue stroma present
5.	Ameloblastic Fibro- Dentinoma	Calcified mass resembling dentin exactly	More disorganized
6.	Odontoameloblastoma	Calcified dentinal structures more than its ameloblastic counterpart	Ameloblastic islands more than its calcified counterpart
7.	Calcifying Epithelial Odontogenic Tumor	Psammomatous calcifications, Amyloid deposition	No psammomatous calcifications or amyloid deposition
8.	Adenomatoid Odontogenic Tumors	Rosettes, Duct like spaces	No rosettes or duct like spaces found
9.	Calcifying Odontogenic Cyst	Ghost cells and dystrophic calcifications	No ghost cells seen

Conclusion

AFO is a mixed odontogenic tumor which is rarely found. It presents as a mixed radiolucency resembling an odontoma. Although it is a benign neoplasm, clinicians should keep in mind that a radiopaque mass in either jaw patients associated with unerupted teeth can be a possibility of rare mixed odontogenic tumor like AFO. This rare case report emphasizes that how important is the histologic diagnosis of any lesion apart from their routine clinical and radiographic diagnosis. All the differential diagnosis should be ruled out before coming to a conclusive diagnosis. Although AFO has a low incidence of malignant transformation, a long term follow up is opined.

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 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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