Ameloblastic Fibro-odontoma of Mandible

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ABSTRACT
The ameloblastic fibro-odontoma is a rare mixed odontogenic tumor. The ameloblastic fibro-odontoma is a well-circumscribed, painless, slow-growing, and expanding tumor with no propensity for bony invasion. Ameloblastic fibro-odontoma is often asymptomatic and usually detected as a result of failure of tooth eruption. It occurs predominantly in children and young adults with no sex predilection and present mostly in posterior segment of the mandible. Many times it presents as a painless swelling, which is the most common clinical sign. Radiologically, ameloblastic fibro-odontoma shows a circumscribed radiolucency, which contains radiopaque foci of various sizes and shapes. Here we present a case of ameloblastic fibro-odontoma in an 18-year-old male patient associated with a hard painless swelling associated with moderate facial asymmetry in the right mandibular region. On histopathological analysis, all the classic features were noted and diagnosis of an ameloblastic fibro-odontoma was made without any obscurity. Diagnosis of ameloblastic fibro-odontoma remains a challenge for oral pathologists. There are many lesions, e.g. ameloblastic fibro-odontoma, odontoameloblastoma immature complex odontoma, calcifying epithelial odontogenic tumor and calcifying odontogenic cyst, which mimic ameloblastic fibro-odontoma clinically and histopathologically. These conditions must be ruled out before making a definitive diagnosis of ameloblastic fibro-odontoma.

Keywords: Diagnosis, Immature complex odontoma, Mixed odontogenic tumor, Odonto-ameloblastoma.


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INTRODUCTION
Ameloblastic fibro-odontoma (AFO) is a benign, slow-growing, expansile epithelial odontogenic tumor with odontogenic mesenchyme. Neivelle, et al described this entity to be a combination of an ameloblastic fibroma and developing complex odontome and approximately 80% lesions are associated with an unerupted tooth. It may inhibit tooth eruption or displace involved teeth, although teeth in the affected area are vital.

Radiographically, the lesion presents with a well-circumscribed, unilocular or multilocular radiolucent area containing various amounts of radiopaque material of irregular size and form. The large areas of calcification make it impossible to radiographically differentiate from complex odontoma. The lesions are usually diagnosed during the first and second decades of life. It occurs with equal frequency in the maxilla and the mandible and with equal frequency in males and females. Histopathologically, the AFO is characterized by islands, strands and cords of odontogenic epithelium immersed in embryonic connective tissue that mimics primitive dental pulp. Formation of osteodentin and enamel is also observed microscopically. Although, ameloblastic fibro-odontoma is a benign lesion, it is developed at a very early age, with rapid growth and destruction of cortical bone. Histologically, the features of AFO are indistinguishable from the odontoameloblastoma, immature complex odontoma, calcifying epithelial odontogenic tumor and calcifying odontogenic cyst. This report describes an ameloblastic fibro-odontoma in an 18-year-old boy with an emphasis on its clinical features, radiographic features, histopathology, differential diagnoses and management.

CASE REPORT
An 18-year-old boy was referred to the Dental OPD at the Institute of Dental Sciences, Bareilly, complaining about a swelling in the right mandibular region. The medical, social and family histories were unremarkable and there was no personal habits related to oral cavity. Extraoral examination showed moderate facial asymmetry on the right side. On palpation, a hard painless swelling was observed in the right mandibular body. Intraoral examination revealed the lesion was a painless, hard swelling, normal colored mucosa, increase in the volume of alveolar ridge and no definition of vestibular fold extending from distal aspect of permanent right mandibular first premolar till the retromolar pad area (Fig. 1).

Panoramic radiography exhibited large and multiple radiopacities enveloped by the radiolucent zone extending from the region of the right permanent first premolar region up to the anterior border of the ramus of the mandible with the presence of an impacted right lower third molar. The lesion contained scattered foci of calcified material coronal...
Fig. 1: Hard swelling of alveolar ridge extending from distal aspect of #44 till the retromolar pad area

Fig. 2: Panoramic radiograph showing multiple large radiopaque lesion enveloped by the radiolucent zone extending from #44 region to the anterior border of the ramus of the mandible. Impacted #48 is also seen.

to the impacted tooth (Fig. 2). Considering the clinical and radiographic findings, a provisional clinical diagnosis of benign odontogenic neoplasm with hard tissue formation was made. The provisional clinical diagnosis of complex odontome was made. Ameloblastic fibro-odontoma, calcifying epithelial odontogenic tumor and calcifying odontogenic cyst were included in differential diagnoses.

Excisional biopsy was performed, and the mass, including the third molar, was submitted for histopathologic diagnosis. On histopathological analysis, the hematoxylin and eosin-stained sections revealed characteristics of both ameloblastic fibroma and odontoma. Microscopic features show odontogenic epithelium arranged in islands, strands and cords interspersed in moderately cellular connective tissue stroma with predominantly spindle-shaped fibroblasts along with some hemorrhagic areas (Fig. 3A). In some areas, the stroma was delicate resembling ectomesenchymal. High-power microscopy revealed epithelial cells producing large hematoxyphilic calcified areas resembling enamel matrix and dentinoid material (Fig. 3B). Odontogenic epithelium arranged in small island, strands and cords interspersed in under high power (Figs 3C and D). No evidence
of malignancy, such as nuclear pleomorphism was observed, and based on clinicopathologic correlation, the diagnosis of ‘Ameloblastic fibro-odontoma’ was made.

**DISCUSSION**

According to WHO classification of odontogenic tumors, ameloblastic fibro-odontoma has traditionally been classified under odontogenic epithelium with odontogenic ectomesenchyme, with or without hard tissue formation. The term ‘epithelial odontogenic tumor with odontogenic mesenchyme’ is becoming more widely accepted these days and avoids potential controversy over the nature of the neoplasia. The term ‘ameloblastic fibro-odontoma’ represents a histologic combination of ameloblastic fibroma and complex odontoma.\(^1,8\)

There has been a lot of discussion in the literature regarding its proper classification. One point of discussion is the discrimination between neoplasm and hamartoma. Philipson et al.\(^{11}\) indicated that the ameloblastic fibro-odontoma has a hamartomatous character but, in contrast, the ameloblastic fibroma has a neoplastic nature. Most authors now agree that ameloblastic fibro-odontoma is a separate entity but it can be histologically indistinguishable from immature complex odontoma. The relative arrangement of the soft tissues and the stage of development of the involved tooth are the useful criteria for diagnosis.\(^{11}\)

The term ‘ameloblastic fibro-odontoma’ appears in the World Health Organization (WHO) classification of odontogenic tumors, whereas ameloblastic odontoma is called odonto-ameloblastoma in the WHO classification.\(^{12,13}\)

In fact, some cases reported in the literature are truly neoplastic and have showed a malignant differentiation. Immune staining with anti-amelogenin sera proved that tumor epithelium and mesenchyme can potentially mimic the full spectrum of phenotypic changes, and the cellular and molecular events that regulate normal odontogenesis most likely operate to a certain extent in the pathogenesis and differentiation of odontogenic tumors.\(^{9}\) Controversy exists regarding the histogenesis of the mixed odontogenic tumors. Cahn and Blum postulated that ameloblastic fibroma is the histologically least differentiated tumor and develops first into a moderately differentiated form, ameloblastic fibro-odontoma, and eventually into complex odontoma. However, the concept that these lesions represent a continuum of differentiation is not widely accepted, and others feel that they are separate pathologic entities. Most now agree that ameloblastic fibro-odontoma exists as a distinct entity, but it can be histologically indistinguishable from immature complex odontoma.\(^{14}\)

Ameloblastic fibro-odontoma is relatively rare. The prevalence among oral biopsies is about 1% and the frequency of ameloblastic fibro-odontoma among odontogenic tumors is reported as 1 to 3%. Daley\(^{12}\) and others investigated the relative incidence of odontogenic tumors in the Canadian population and found that 3.06% of all odontogenic tumors were ameloblastic fibro-odontomas. Ameloblastic fibro-odontoma usually occurs in people less than 20 years old, and age is thus an important characteristic in the differential diagnosis.\(^{2,8}\) Hooker reported the mean age of patients as 11.5 years (6 months-39 years).\(^8\) In presently described case, in which the patient was 18 years old, agrees with the above observations regarding age range as described by different authors. There is no difference in prevalence between the sexes.\(^7,8\) In the present case, the patient was a boy.

Ameloblastic fibro-odontoma is usually found in the molar area. The distribution is roughly equal between the maxilla and mandible.\(^7,8\) However, in the present case, the lesion was present with respect to right posterior mandible.

The two most common presenting complaints are swelling and failure of tooth eruption. The lesion may displace the erupted teeth, but other symptoms, such as pain and paresthesia, are uncommon. Asymptomatic cases are usually discovered incidentally on radiography. This lesion is generally considered a slow-growing central jaw tumor; however, several exceptions to this pattern have been reported.\(^1\) Occasionally, the tumor exhibits marked swelling, which results in facial disfigurement.\(^9\) Finally, an AFO sometimes inhibits tooth eruption.\(^13\) In the present case also, the significant facial disfigurement was observed because of swelling, representing its sprawling nature.

Radiography usually shows a well-defined radiolucent area containing various amounts of radiopaque material of irregular size and form.\(^{16}\) The ratio of radiopaque to radiolucent areas differs from one lesion to another; sometimes the mineralized element in the tumor predominates and the lesion may resemble an odontoma.\(^{17}\) Some of the lesions are relatively small when first detected, measuring 1 to 2 cm in diameter, whereas others may be exceedingly large, involving a considerable portion of the body of the mandible or maxilla.\(^19\) In the present case, large and multiple radiopacities enveloped by the radiolucent zone extending from the region of the right permanent first premolar region up to the anterior border of the ramus of the mandible with the presence of an impacted right lower third molar, which is consistent with the findings of other authors.

The other lesions which are showing mixed radiolucent and radiopaque patterns are calcifying epithelial odontogenic tumor, calcifying odontogenic cyst, immature complex odontoma and possibly adenomatoid odontogenic tumor.\(^{19}\)

Histopathologically, the relative arrangement of the soft tissues and the stage of development of the involved tooth are useful criteria for diagnosis.\(^1\) The tumor mass is surrounded by a fibrous capsule and is composed predomi-
nantly of a fibroblastic connective tissue matrix containing strands of odontogenic epithelium and immature tooth structures, including enamel and dentin. The connective tissue is moderately cellular with spindle-shaped fibroblasts. No evidence of malignancy is found. As mentioned above, the diagnosis of present case was also made, considering the same histopathologic criteria.

The histological characteristics of AFO cannot be distinguished from odontoma, and consequently, some authors believe that ameloblastic fibro-odontoma is an immature complex odontoma. The large amount of calcifying tissue present in the lesion may speak in favor of the theory that AFO is an early stage of odontoma. In the same way, no ultrastructural differences were observed between AFO and ameloblastic fibroma but, histologically, thin strands or cords of epithelium that resembles dental lamina and cap and bell stage of early odontogenesis can be seen in ameloblastic fibroma. Ameloblastic fibro-odontoma can be differentiated from calcifying odontogenic cyst on the basis that it is a well-circumscribed, solid or cystic lesion derived from odontogenic epithelium that microscopically resembles ameloblastoma but differs by containing ghost cells and spherical calcifications. Calcifying epithelial odontogenic tumor microscopically consists of sheets and strands of polyhedral epithelial cells, homogenous eosinophilic deposits that stain positive for amyloid, and spherical calcifications which differentiates it from ameloblastic fibro-odontoma. In odontoameloblastoma, there is presence of sheets of typical ameloblastoma of recognized types, usually basal cell, follicular or plexiform which differentiates it from ameloblastic fibro-odontoma and tumor cells recapitulate stratum intermedium layer of early bell stage of odontogenesis.

The recommended treatment for ameloblastic fibro-odontoma is conservative surgery with enucleation because it is a well-circumscribed benign tumor and there is little tendency to local invasion. When the lesion includes an unerupted tooth, the tooth should be removed with the mass. There is very little potential for recurrence. Malignant transformation of ameloblastic fibro-odontoma is rare, and its exact rate is not known. Howell and Burkes reported two cases of ameloblastic fibro-odontoma that showed malignant transformation to ameloblastic fibrosarcoma. The present case was treated with conservative surgery with enucleation along with removal of impacted third molar tooth. After a follow-up of 9 months, no recurrence was observed and the healing was without any complications.

In conclusion, the case reported here represents an ameloblastic fibro-odontoma causing significant facial asymmetry with jaw expansion. Diagnosis of ameloblastic fibro-odontoma remains a challenge for oral pathologists. There are many lesions which mimic ameloblastic fibro-odontoma clinically and histopathologically. These conditions must be ruled out before making a definitive diagnosis of ameloblastic fibro-odontoma.

REFERENCES