Case Report

Ameloblastic Fibro-Odontoma - An Insight into its Clinico-Histopathological, Radiographic Features and its Surgical Management

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ABSTRACT:

Ameloblastic fibro-odontoma (AFO) is a rare, benign mixed odontogenic tumor characterized by the presence of epithelial tissue, mesenchymal tissue and dental hard tissues such as enamel and dentin. The lesion is more frequently observed in children and young adults. Clinically, AFO manifests as a painless swelling in the jaw, often associated with impacted or unerupted teeth. Radiographically, it appears as a well-defined, mixed radiolucent-radiopaque lesion. Histologically, it resembles ameloblastic fibroma (AF) with additive formation of enamel and dentin. Treatment generally involves surgical enucleation, with recurrence being rare. We present a case report of a 17 year old female with painless swelling on the left side of mandible, diagnosed with AFO and managed surgically by enucleation.

KEY WORDS: Ameloblastic fibro- odontoma, ameloblastic fibroma, complex odontoma, mixed odontogenic tumors, odontoma

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INTRODUCTION:

Ameloblastic fibro-odontoma (AFO) is an uncommon, benign mixed odontogenic tumor.[1] WHO in 2005 has defined it as a neoplasm composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles dental hard tissues like enamel and dentin.[1] The term AFO was first coined by Hooker in 1967.[2] Prior to 1967 the AFO was confused with ameloblastic-odontoma (odontoameloblastoma) which was aggressive in nature and shared biological behavior with ameloblastoma but in 1967 Hooker distinguished between the two and found it to be less aggressive.[1] The pathology is frequently seen in children between 5-14 years predominantly in molar region.[1,3] Clinical presentation may vary from asymptomatic to symptoms like swelling, delay in eruption of tooth or displacement of teeth and cortical expansion.[4] Radiographically, AFO displays a well-demarcated and encapsulated lesion filled with varying amounts of irregular and haphazardly arranged radiopaque mass mimicking complex odontoma.[2] Histologically it resembles ameloblastic fibroma (AF) with additive formation of enamel and dentin.[2] Due to its benign nature, AFO generally responds well to conservative surgical excision, with a low recurrence rate. This case report demonstrates clinicohistopathological radiographic features of AFO with its management.

CASE REPORT:

A 17 year old female reported to the Department of OMFS, of our institute with chief complaint of painless swelling in her lower left back region of jaws for 6 months. There was no family

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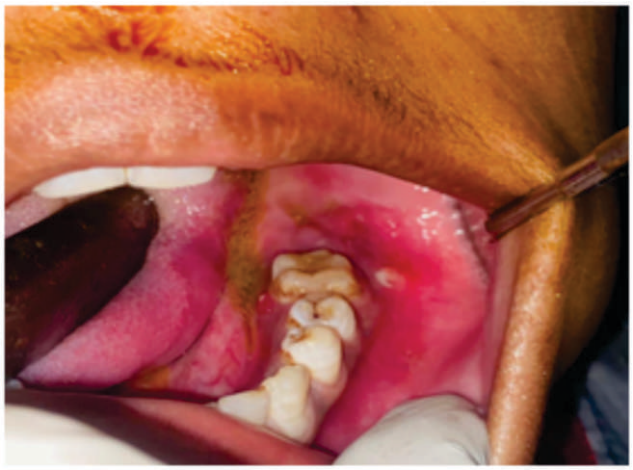

Figure 1: Intraoral picture showing vestibular obliteration.

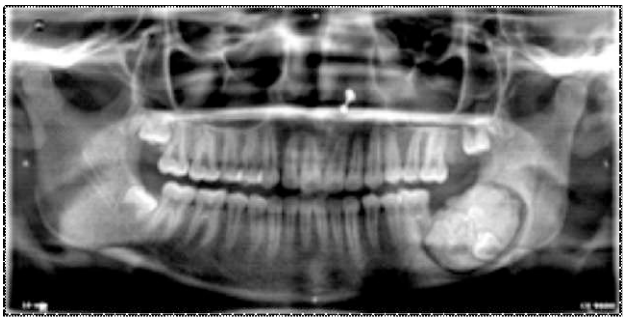

Figure 2: OPG reveals mixed radiopaque, radiolucent mass in the third molar region with unerupted second molar.

history and no past history of trauma or infection. On extraoral examination, facial asymmetry could be noted because of single diffuse swelling 3 cm away from the corner of mouth to the angle of mandible in anterior-posterior direction and superior-inferiorly from the ala tragus line to 1 cm below the lower border of mandible. On intraoral inspection, vestibular obliteration was noticed in the 36, 37, 38 region (Figure 1). Mucosa over the swelling appeared reddish in colour. Missing 37 and 38 were observed. On palpation, swelling was non tender, firm and bony hard in consistency. OPG revealed a well-defined radiopaque lesion extending from distal root of the right 1st molar to the ramus of mandible and an unerupted 37 tooth was observed inside the mass (Figure 2). Root resorption of 36 was evident. CBCT revealed well encapsulated radiopaque lesion measuring 32.1mm x 24.3 mm with expansion of both the buccal and lingual cortical plate and no evidence of cortical plate perforation (Figure 3).

The provisional diagnosis of complex odontoma was made. Differential diagnosis of AFO, calcifying odontogenic cyst, pindborg tumour and odontoamelo-blastoma were made based on the clinical and radiographic presentation.

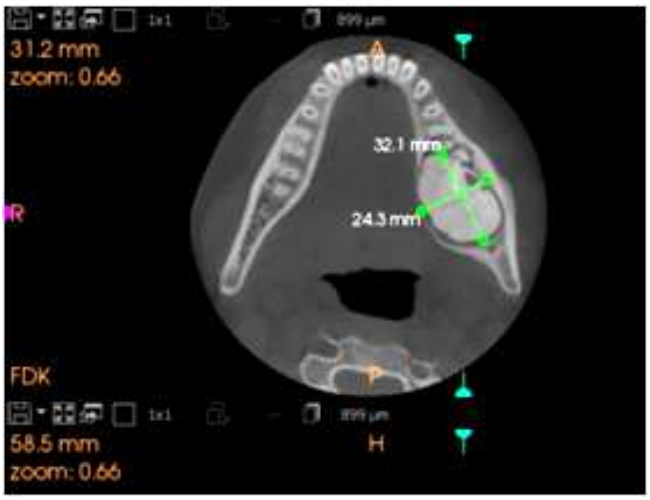

Figure 3: CBCT revealing well encapsulated lesion with expanded cortices.

The pathological and biochemistry reports (PTH, serum alkaline phosphatase, serum calcium) were within normal range. Considering her age, well encapsulation of the lesion nature and normal biochemistry results, we opted for a conservative enucleation of the lesion.

Under general anesthesia following aseptic protocol, crevicular incision was placed from 34 to 38 region with a vertical relieving incision on the retro molar region. The calcified mass was exposed after elevating the full thickness mucoperiosteal flap. The calcified mass was sectioned into fragments and removed along with the unerupted 37 (Figure 4). The lesion was completely excised and the residual defect margins were smoothened. The specimen was sent for histopathological examination. The inferior alveolar nerve was intact without causing any injury.

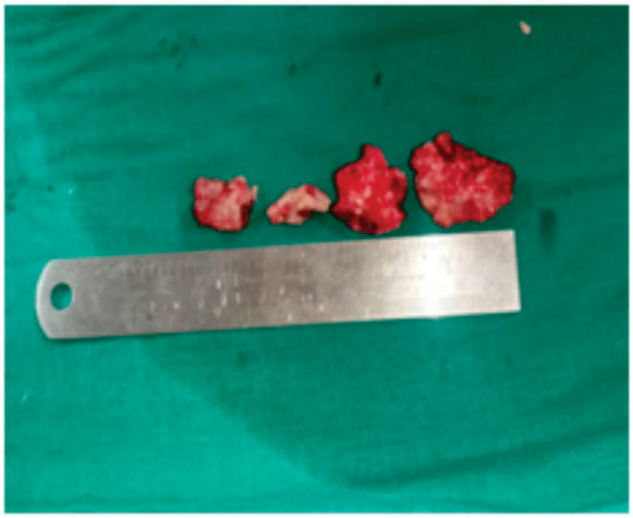

Figure 4: Extracted mandibular left second molar tooth with removal of calcified mass.

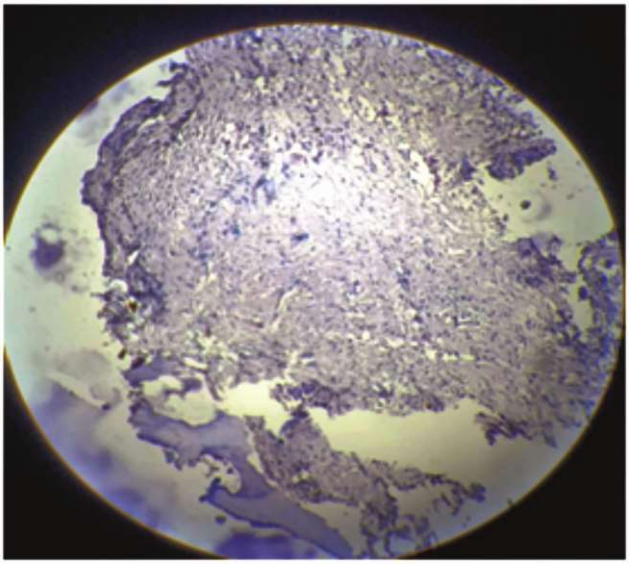

Figure 5: Histopathology of Ameloblastic fibro-odontome.

Histopathology showed strands, cords and islands of odontogenic epithelial cells with central stellate reticulum-like cells (Figure 5). Some of the follicles showed peripheral tall columnar cells with central stellate reticulum-like cells. The connective tissue was delicate and composed of thin collagen fibres and stellate shaped fibroblasts with areas resembling dental papilla-like tissue. The lesion revealed abundant eosinophilic material resembling mature dentine and dentinoid like areas along with enamel matrix, basophilic spherules resembling cementum. Peripherally parallel bundles of collagen was seen. Based upon the histopathological report, a final diagnosis of AFO was made.

The patient was reviewed after a week for suture removal with uneventful recovery and healing. There was no complaint of paresthesia and no sign of nerve injury. The patient was called for regular follow up till 6 months. No complications were noted.

DISCUSSION:

AFO is a rare mixed neoplasm of odontogenic epithelium with ectomesenchymal tissue with dental hard tissue formation.[1] The incidence of AFO varies from 0.3% to 1.7% among the odontogenic tumours, and 71 % only between 5 to 14 years with mean age of 9.4 years so it is also called as tumour of childhood.[1] AFO lesions have equal preponderance to mandibular and maxillary region with higher occurrence in posterior region. Males are affected more as compared to female.[5] Clinically, AFOs are asymptomatic and so the patient does not seek any consultation till the lesion becomes large in size causing facial disfigurement and bony expansion.[6] Delayed or unerupted tooth may also be one of the concern.[4] The unerupted or displaced tooth in apical direction obviates the origin of the lesion from tissue of dental origin primarily dental lamina.[5] In our case, the presenting feature was painless swelling in a teenage girl. As the swelling was painless, the consultation was done only when the swelling attained a size to be visualized externally. The delay in consultation may be because of the painless nature of the lesion, ignorance of patients belonging to uneducated farmer background, poor socioeconomic status and scarcity of experienced dental surgeons in rural areas.[7]

The exact histogenesis of AFO is controversial. Different studies have different opinions. Cahn and Blum[8] postulated the concept of continuum of differentiation i.e. least differentiated tumor gets transformed into well differentiated tumor. The origin of AFO is from Ameloblastic Fibroma (AF) which is a least differentiated tumour that gets transformed into a moderately differentiated form which is AFO and finally AFO into Complex Odontoma. But this concept was rejected by Slootweg et al[9] who found that AF and AFO are separate entity because:

a) The AFO which is a more differentiated form appears early in life than AF.
b) The site predilection of AFO is different from AF.
c) AF has neoplastic character but AFO has hamartomatous features.

Radiographically, AFO presents as well-demarcated circumscribed unilocular radiolucencies with variable amounts of calcifications.[6] In the present case we also observed the similar finding of a well - defined mixed lesion extending from distal root of the right 1st molar to the ramus of mandible and an unerupted 37 tooth was observed inside the mass.

AFO has been classified under developing odontoma in WHO classification (2017 and 2022) and are considered hamartomas rather than tumour but the presence of BRAF p.V600E mutation in AFO which is similar to that of AF and is absent in odontoma favours the argument that some lesions are neoplastic which manifest aggressive biological behavior (displacement and/or prevent eruption of teeth and continued rapid growth with cortical perforation) large size and reoccurrence.[4,10]

Histopathological section show islands and strands of odontogenic epithelium lined by tall columnar cells resembling ameloblasts and central stellate reticulum-like tissue scattered within a highly cellular connective tissue resembling primitive dental papilla. Decalcified H&E stained sections show

conglomerate mass of enamel and dentine arranged in a disorganized form .[4] In our case, the decalcified H&E stained section showed strands, cords and a few islands/ follicles of odontogenic epithelial cells. Some of the follicles show peripheral tall columnar cells with central stellate reticulum-like cells. The connective tissue was delicate and composed of thin collagen fibres and stellate shaped fibroblasts with areas resembling dental papilla-like tissue. The centre of the lesion showed abundant of eosionophilic material resembling mature dentine and dentinoid like areas along with enamel matrix, basophilic sperules resembling cementum. Abundant collagen fibres were seen in periphery.

The differential diagnosis include mixed odontogenic tumour like calcifying odontogenic cyst, calcifying odontogenic tumor, immature complex odontoma and odontoamelo-blastoma. AFO can be differentiated from calcifying epithelial odontogenic cyst by the presence of ghost cells within epithelial lining and calcifications.[4] In calcifying epithelial odontogenic tumor presence of liesegang ring is seen.[4] The term odontoamelo-blastoma exhibit histological features similar to ameloblastoma and complex odontoma.[4]

The recommended management for AFO is enucleation as the tumor is firmly encapsulated and has no tendency to invade the surrounding tissue. AFO is thought to be less aggressive and is effectively treated with enucleation with removal of the unerupted tooth.[1,2,5,8] Recurrence of 7.4% has been reported in literature which was attributed to failure to remove the lesion completely or unerupted or displaced tooth.[1] Malignant transformation of AFO is rare so long term follow up is essential.[11]

In the present case although the size of the lesion was more than 3 cm but lesion was well encapsulated without any local invasion curtailing the enucleation with removal of unerupted 37 and also extraction of 36 which had undergone external root resorption.

CONCLUSION:

AFO is a rare, benign mixed odontogenic tumor with nonspecific clinical and radiographic features. Histopathology can only confirm the diagnosis. Although the incidence of the lesion is less than 2 percent, it should always be considered in differential diagnosis of the mixed radiolucent radiopaque lesion in paediatric age groups. Owing to its well encapsulation, non-invasive nature, low aggressiveness and low recurrence, it is managed conservatively by enucleation with extraction of unerupted teeth. Long term follow up is mandated to know any malignant transformation.

DECLARATION OF PATIENT CONSENT:

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s)/guardian has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients/guardian 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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