A Rare Case of Ameloblastic Fibro-Odontoma of Mandible with Literature Review

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Abstract

Ameloblastic fibro-odontoma (AFO) is a benign, mixed odontogenic tumor of epithelial and mesenchymal origin. It is predominantly seen in the younger age group, with delayed or altered eruption of teeth. Being clinically asymptomatic, this is identified incidentally during routine radiographic examination. Although considered to be nonaggressive with minimal tendency for recurrence, AFO requires surgical excision with long-term follow-up. This paper presents a rare case report of an 11-year-old boy, who was provisionally diagnosed with complex odontoma and later turned out to be AFO of the mandible.

Keywords: Ameloblastic, fibro-odontoma**,** odontogenic tumors

Introduction

Odontogenic tumors are heterogeneous group of lesions originating from the cells or tissues of the tooth-forming apparatus, which can be epithelial or mesenchymal, or both (mixed) (Thoma KH and Goldman HM).[1] Odontogenic tumors exhibit specific histopathological characteristics pertaining to various stages of odontogenesis with diverse clinical manifestations. The biological behavior of these lesions can be hamartomatous in nature or slow-growing benign neoplasms to aggressive malignant tumor.[2] Ameloblastic fibro-odontoma (AFO) is a benign, mixed odontogenic tumor of the jaws. This pathological entity has been classified by the World Health Organization (WHO) under the odontogenic epithelium with odontogenic ectomesenchyme with or without hard-tissue formation.[3] AFO is histologically composed of three predominant elements (1) immature fibroblastic connective tissue, (2) ectodermal component, and (3) mineralized component.[4,5] Even till date, there is a lack of consensus amid mixed odontogenic tumors to state that AFO is a distinct entity. It is believed that these tumors represent in different stages of maturation. Mixed odontogenic tumors present as a challenge to the clinicians, because nonodontogenic lesions such as immature odontomas and calcifying epithelial odontogenic cyst present clinically alike to odontogenic tumors. Differential diagnosis should include lesions with mixed radiographic patterns, such as calcifying epithelial odontogenic tumor, calcifying odontogenic cyst, and adenomatoid odontogenic tumor.

Case Report

An 11-year-old boy reported to our institution with complaints of swelling in the left lower border of the mandible measuring 3 cm × 3.5 cm, extending 5 cm from the corner of the mouth to the ramus posteriorly and superiorly till the imaginary line drawn from the tragus to the corner of the mouth and inferiorly involving the lower border of the mandible. The patient was advised for an orthopantomogram (OPG) and computed tomography (CT) of the involved region. OPG revealed an irregular well-defined radiolucent area with mixed radiopaque mass in relation to the lower border of the mandible, measuring 2 cm × 2.5 cm [Figure 1]. CT scan revealed no perforation of the lingual cortex. Surgical excision of the calcified mass was planned and surgical consent was obtained.

Under endotracheal intubation, crevicular incision was placed from 38 to 33 with a vertical releasing incision; the calcified

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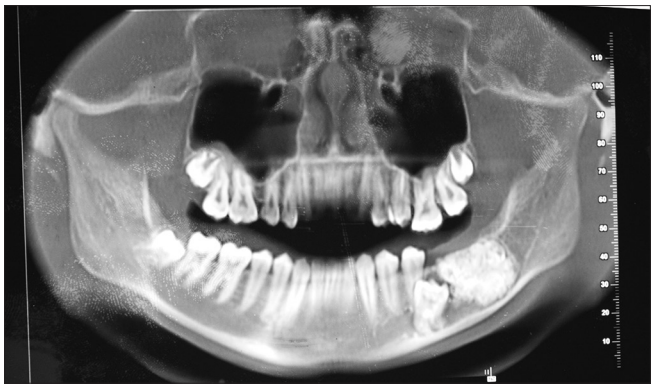

Figure 1: Orthopantomogram reveals mixed radiopaque, radiolucent mass in the third molar region with impacted first molar

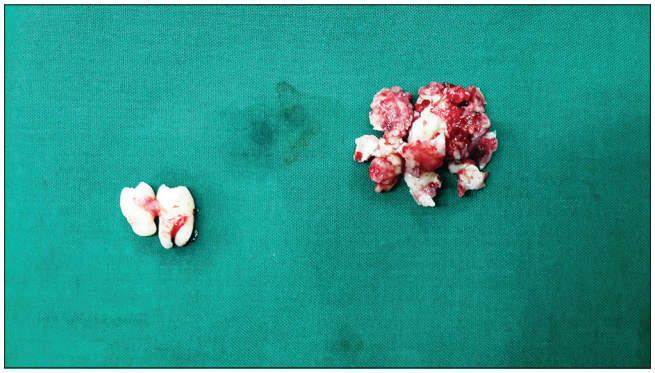

Figure 3: Extracted teeth and specimen

mass along with the impacted third molar was exposed after elevating the full-thickness mucoperiosteal flap. The bulkiness of the calcified mass was sectioned into fragments and removed in portions [Figure 2]. Complete excision was not attempted in lieu with lingual plate to avoid pathological fracture of the jaws. The lesion was excised, and the residual defect margin was smoothened [Figure 3]. Specimen was sent for histopathological examination. The impacted third molar was removed without causing injury to the inferior alveolar nerve. The nerve was intact and displaced inferiorly below the calcified mass.

The patient reviewed after a week for suture removal with uneventful recovery and healing. The patient was asymptomatic with no complaints of nerve morbidity. The patient was called for a regular follow-up.

Discussion

AFO is a tumor of mixed odontogenic origin which constitutes 1%–3% of all odontogenic tumors. At present, it is widely recognized and accepted as an epithelial odontogenic tumor with odontogenic ectomesenchyme with or without hard-tissue formation.[3]

AFO is an uncommon odontogenic tumor, with relative occurrence in the earlier stages (first two decades, mean age: 11.5 years) of life, in comparison with other mixed odontogenic tumors

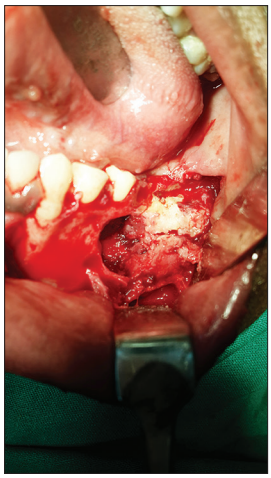

Figure 2: Intraoperative exposure of the calcified mass

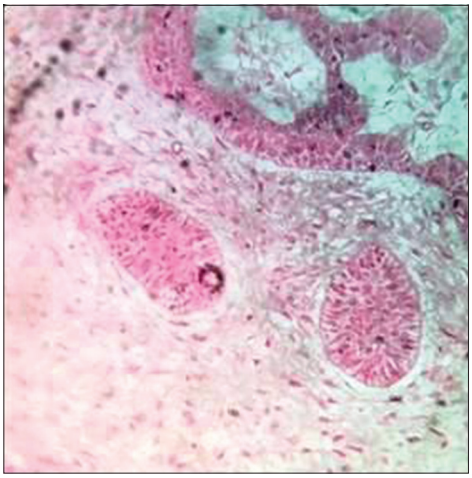

Figure 4: High magnification showing odontogenic epithelium

which makes age a critical benchmark in considering AFO in the differential diagnosis of posterior radio-opaque lesions of the jaws.[4,5] AFO can also occur at advanced ages.[6] The tumor occurs equally in the maxilla and mandible, predominantly in the molar region with no specific gender predilection, slightly with a male preponderance.[7]

Clinically, the lesion presents with swelling and failure of tooth eruption. The associated tooth is displaced in an apical direction. The asymptomatic swelling is likely to be seen in the posterior portion of the maxilla or mandible and failure or delayed eruption of the tooth involved. Paresthesia is not evident in cases of AFO. Secondary infection of AFO is also relatively uncommon.

There has been a great consensus in classifying mixed odontogenic tumors, ameloblastic fibroma (AF), AFO, mature and immature compound, and complex odontomas. Some authors believe that they are separate entities while others consider that they represent different stages of the same lesion and the extent of development determining the histologic differences.

Radiographically, it presents as a well-defined radiolucent area with mixed radiological material resembling different odontogenic neoplastic formations, with varied size and forms. However, the final diagnosis is confirmed upon histological evaluation, where AFO microscopically exhibits as strands, cords, and islands of proliferating odontogenic epithelium enclosed by cellular ectomesenchymal tissues that resemble the primitive dental pulp (dental papilla), dental lamina, and enamel organ. It also exhibits varied levels of inductive changes and dental hard-tissue formation. This feature of enamel matrix production (amelogenin) makes it exclusive, microscopically in differentiating AFO from AF, ameloblastic fibrodentinoma, and mature complex odontomas.

The histogenetic order of differentiation of mixed odontogenic tumors is mostly debatable. AFO is histologically seen as a proliferation of odontogenic mesenchyme resembling dental papilla such as stromal cells. Cahn and Blum (1952) postulated that AF, the least differentiated tumor, develops first into a moderately differentiated form, AFO, and sooner or later into a complex odontoma. However, the concept that these lesions represent a continuum of differentiation is not widely accepted, and others believe that they are separate pathological entities.

Histologically, according to the criteria of the WHO, the tumor consists of the same type of epithelial and mesodermal tissue elements as observed in the AF, however, in combination with calcified odontogenic tissue composed predominantly of a fibroblastic connective tissue matrix containing strands of odontogenic epithelium and immature tooth structures, including enamel and dentine [Figure 4].

The management of AFO is limited to conservative surgical approach by enucleation as the tumor is well encapsulated with very little propensity to local invasion. AFO has been considered to be less aggressive and can be treated adequately by enucleation and removal of the involved tooth structure. Disputes regarding tooth removal exist though there are case reports and literature evidence favoring both removal and preservation. There is very little evidence in literature depicting the recurrence of the tumor from leftover remnants of the lesion.

Despite the lesions' small potential for recurrence, recurrences have been attributed to the inadequate surgical removal of the lesion at the time of primary surgical management which mandates follow-up.

Malignant transformation of AFO is rare but has been reported to occur at a relatively older age.[8] Hence, long-term follow-up is required. They occur frequently than AF and Ameloblastic odontomas.

Conclusion

The conferred case clinically, radiographically, and histologically is AFO which is a rare benign, mixed odontogenic tumor. Treatment of this lesion is mostly excision, since most of them respond to conservative management. It should always be considered during differential diagnosis of the mixed radiopaque lesions in the intraoral region, especially with patients of younger age group.

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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Conflicts of interest

There are no conflicts of interest.

References

1. Thoma KH, Goldman HM. Odontogenic tumors: A Classification based on observations of the epithelial, mesenchymal, and mixed varieties. Am J Pathol 1946;22:433-71.
2. Niranjan KC, Shaikh Z. Clinicopathological correlation of odontogenic cysts and tumours in a South Indian population over a 20-year period. Int J Dent Res 2014;2:32-6.
3. Barnes L. World Health Organization Classification of Tumours. Pathology and Genetics of Head and Neck Tumours. International Agency for Research on Cancer (IARC): Lyon, France; 2005.
4. Slootweg PJ. An analysis of the interrelationship of the mixed odontogenic tumors-ameloblastic fibroma, ameloblastic fibro-odontoma, and the odontomas. Oral Surg Oral Med Oral Pathol 1981;51:266-76.
5. Reichart PA, Philipsen HP, Gelderblom HR, Stratmann U. Ameloblastic fibro-odontoma-report of two cases with ultrastructural study of tumour dental hard structures. Oral Oncol Extra 2004;40:8-12.
6. Kumar LK, Manuel S, Khalam SA, Venugopal K, Sivakumar TT, Issac J. Case report open access international journal of surgery case reports ameloblastic fibro-odontoma. Int J Surg Case Rep 2014;5:1142-4.
7. Buchner A, Kaffe I, Vered M. Clinical and radiological profile of ameloblastic fibro-odontoma: An update on an uncommon odontogenic tumor based on a critical analysis of 114 cases. Head Neck Pathol 2013;7:54-63.
8. Howell RM, Burkes EJ Jr. Malignant transformation of ameloblastic fibro-odontoma to ameloblastic fibrosarcoma. Oral Surg Oral Med Oral Pathol 1977;43:391-401.