Case Report

Ameloblastic fibrodentinoma of the anterior mandible: A rare case report
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Abstract
Odontogenic tumors constitute a heterogeneous group of lesions with diverse histopathologic features and clinical manifestations. Ameloblastic fibrodentinoma (AFD) is a rare benign odontogenic tumor and is thought to be a spectrum of AF and ameloblastic fibro-odontoma. AFD presents as slow-growing, asymptomatic lesion commonly associated with an unerupted tooth in the mandibular posterior region. If a lesion involves deciduous dentition, it occurs in the anterior jaw. It is commonly seen in males than in females. It may present as intraosseous or extraosseous variants.

Keywords: Ameloblastic fibrodentinoma, Anterior mandible, Permanent teeth

Introduction
Ameloblastic fibrodentinoma (AFD) was first discovered by Strait in 1936 and named it as dentinoma.[1] Later, it was called as immature dentinoma, AF with dentinoid formation, and fibro ameloblastic type of dentinoma by various authors.[2] Different classifications have been applied to this tumor. Earlier to the 1992 WHO classification, the term AFD was commonly used. In the 1992 WHO classification, AFD is considered neoplasm similar to AF that shows inductive changes leading to the formation of dentin. Consequently, the terms AFD and dentinoma were used interchangeably in this classification. However, the existence of dentinoma as a separate entity has to be proved.[3]

AFD presents as slow-growing, asymptomatic lesion commonly associated with an unerupted tooth in the mandibular posterior region. If a lesion involves deciduous dentition, it occurs in the anterior jaw. It is commonly seen in males than in females.[4] It may present as intraosseous or extraosseous variants.[5] Here we report a rare case of AFD in 32 year old male occurring in the anterior region of the mandible in association with permanent teeth.

Case Report
A 32-year-old male patient reported to the Rajarajeswari Dental College and Hospital, Bengaluru, for the evaluation of swelling in the lower anterior region of the jaw. A history revealed the appearance of an asymptomatic swelling in the lower anterior region of jaw for 6 months which gradually increased to present size. Extraoral examination revealed a diffuse swelling in the anterior region of the mandible. Intraoral examination revealed non-tender, well-defined, solitary, bony hard swelling extending from the left mandibular permanent canine to the right mandibular permanent canine measuring about 4 cm × 3 cm, thereby obliterating the buccal vestibule. Associated teeth showed Grade I mobility. However, overlying mucosa was pink in color and appeared to be stretched. Adjacent gingiva appeared normal [Figure 1]. No history of trauma was given, and the medical history was non contributory.

Imaging examination
The patient was subjected to radiological examination which included orthopantamograph (OPG) and computed tomography (CT) imaging. OPG revealed irregular radiolucent lesion with respect to 32–42 regions with specks of radiopacity. Roots of 31 and 41 were displaced [Figure 2]. CT revealed well-defined radiolucent lesion displacing the roots of 31 and 41 lingually and extending from 32 to 44 regions. Destruction of the labial cortical plate is also evident in the same region [Figure 3].

Based on clinical and radiological features, a provisional diagnosis of odontogenic tumor was rendered. Incisional biopsy was performed.
**Histopathological findings**

The histopathological examination of the specimen revealed neoplastic proliferation of multiple islands and strands of odontogenic epithelium in the cell-rich primitive ectomesenchyme. Myxoid degeneration was also evident surrounding the odontogenic epithelium [Figure 4]. Odontogenic islands encompassed of peripheral tall columnar, ameloblast-like cells with reversal of polarity and central stellate reticulum-like cells. Focal areas in the stroma revealed juxtaepithelial homogeneous hyalinization resembling dentinoid matrix [Figure 5]. Cystic degeneration was seen in few odontogenic islands [Figure 6]. The stroma was highly cellular with numerous plump fibroblasts resembling dental papilla. Atypia was not seen in epithelial or mesenchymal cells. Enamel matrix was not appreciated in any of the histological sections. Based on the above-mentioned microscopic features, the diagnosis of AFD was made. Further, wide surgical excision was done along with extraction of 31 and 41 as our case showed.

![Figure 1: Pre-operative intraoral photograph shows well-defined swelling extending from 33 to 43](image1)

![Figure 2: Panoramic radiography shows ill-defined radiolucent lesion with specks of radiopacity with respect to 32–42](image2)

![Figure 3: Horizontal computed tomography revealed well-defined radiolucent lesion from 32 to 44 regions. Destruction of labial cortical plate is also noted in the same region](image3)

![Figure 4: Photomicrograph showing strands and islands of odontogenic epithelium in a cell-rich ectomesenchyme with myxoid degeneration around odontogenic epithelium (×4)](image4)

![Figure 5: Photomicrograph showing odontogenic islands made of peripheral ameloblast-like cells with reversal of polarity and central stellate reticulum-like cells. Dentinoid like material adjacent to the odontogenic epithelium is also noted (×20)](image5)
aggressive feature such as labial cortical perforation. The gross specimen was well encapsulated measuring about 2 cm × 3 cm in size. The diagnosis of AFD was confirmed on excisional biopsy.

Discussion

AFD is a rare, mixed odontogenic tumor composed of odontogenic epithelium and odontogenic ectomesenchyme with dentine or dentine-like tissue formation. In the WHO (2005) classification of odontogenic tumors, both AFO and AFD have been considered as distinct entities even though they are clinically and pathologically almost same. The presence of enamel and dentine together or only dentine helps in the delineation of these two lesions.

AFD presents as slow-growing, asymptomatic lesion commonly associated with unerupted tooth in the mandibular posterior region. If a lesion involves deciduous dentition, it occurs in the anterior jaw region. Along with central lesion, peripheral or gingival lesions are also noted which is reported exceedingly rare. In our case, tumor occurred within lower anterior region of jaw but in relation to permanent teeth.

Histologically, AFD is composed of proliferating odontogenic epithelium in the form of strands and islands in cellular ectomesenchymal tissue which resembles dental papilla. Odontogenic islands are composed of tall columnar to cuboidal ameloblast-like cells with reverse nuclear polarity and central stellate reticulum-like cells. Proliferating epithelium also shows varying degrees of inductive effects on the mesenchyme, leading to the formation of varying amounts of dentine. Similar features were noted in our case also.

Treatment and prognosis

Simple enucleation remains the treatment of choice for isolated lesions of the jaw. A conservative approach remains recommended for AFD. Since AFD is a benign lesion, the recurrence rate is very less and very rarely the ameloblastic fibrodentinosarcoma, and malignant odontogenic neoplasm is thought to result from the malignant transformation of the ectomesenchyme of AFD.

Conclusion

Here, we present a rare case of AFD in the anterior region of the mandible in relation to permanent teeth. A proper clinical, radiological, and histological evaluation must be established and the treatment should be planned accordingly for AFD cases. A regular follow-up is required to rule out any evidence of recurrence and malignant transformation.

References
