

CASE REPORT



Ameloblastic fibrodentinoma: A rare occurrence

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Received 02 February 2016;

Accepted 05 March 2016

doi: 10.15713/ins.ijmdcr.44

How to cite the article:

Shwetha V, Vineeth K, Prasad K, Mysorekar V, Rajanikanth BR. Ameloblastic fibrodentinoma: A rare occurrence. *Int J Med Dent Case Rep* 2016;2:1-4.

Abstract

Ameloblastic fibrodentinoma (AFD) is a rare tumor belonging to a group of lesions referred to as mixed odontogenic tumors. Mixed odontogenic tumors are neoplasms consisting of proliferating odontogenic epithelium embedded in a cellular ectomesenchyme which closely resembles dental papilla and has varying degrees of inductive change and hard tissue formation. These lesions range in biologic behaviors between hamartomas and true neoplasms. This group of lesions includes ameloblastic fibroma, AFD, and ameloblastic fibro-odontoma. A rare case of AFD manifesting in the mandible is reported.

Keywords: Ameloblastic fibrodentinoma, mandible, mixed odontogenic tumor

Introduction

Odontogenic tumors are a heterogeneous group of diseases with diverse clinical and histopathological features ranging in biologic behaviors from hamartomas to benign and malignant neoplasms with metastatic potential.^[1,2] Odontogenic tumors originate from the odontogenic epithelium, ectomesenchyme, and mesenchyme.^[2] Ameloblastic fibrodentinoma (AFD) is a rare odontogenic tumor composed of odontogenic epithelium and odontogenic mesenchyme with dentin or dentin-like tissue and is hence referred to as a mixed odontogenic tumor.^[1]

AFD is also known as dentinoma or fibro-ameloblastic dentinoma.^[3] The first case of AFD was reported by Straith in 1936.^[2] As this tumor is rare and difficult to interpret, the related nomenclature has been subject to modifications during these years. According to the 1992 WHO classification, this tumor is named as "AFD."^[3,4]

This tumor occurs commonly in the younger age group, below 30 years. Mandible is the most commonly affected site when compared to maxilla (maxilla:mandible - 1:3).^[1,3,4] It has been reported that when this tumor is associated with deciduous teeth, it is generally located in the incisor region while those occurring in and around the permanent teeth show a predilection for the molar region.^[3,4]

Radiographically, it manifests as a mixed radioopaque and radiolucent lesion showing a fairly well-delineated radiolucency

with varying degrees of radiopacity owing to the formation of dentin. The recommended treatment of AFD is surgical excision.^[2]

Current information on the incidence of this lesion comes from sporadic case reports. A case of AFD manifesting in the mandible has been reported.

Case Report

A 27-year-old male patient reported to us with the chief complaint of pain and inability to open his mouth since 1 week. The patient was apparently well 3 months ago when he initially experienced pain and swelling in his mandibular right molar region. He had visited many other dental practitioners and was prescribed antibiotics and analgesics. However, the patient used to have a pain free period for a very short span after taking medications following which he experienced recurrence of the pain and restriction in mouth opening. Presently, he reported a similar recurrence since 3 days associated with pain, swelling, and inability to open his mouth.

Clinical examination revealed a limited mouth opening with a diffuse extraoral swelling on the right lower third of the face extending superoinferiorly from the line joining the angle of the mouth and the tragus to the inferior border of the mandible and anteroposteriorly from 3 cm behind the angle of the mouth to

the angle of mandible. Intraoral examination revealed a swelling obliterating the vestibule in the right mandibular posterior region measuring approximately 2 cm × 3 cm in size. The overlying mucosa was inflamed with pus discharge from the distal gingival sulcus in the 46 region. On palpation swelling was firm to hard in consistency, tender with buccal cortical plate expansion in relation to 47 and 48 region. Hard tissue examination revealed a missing 47 and 48. No other significant findings other than missing 36 and a grossly decayed 16 were noted. A provisional diagnosis of right submasseteric space infection secondary to a pericoronal abscess in relation to 46 region was made. A differential diagnosis of an infected dentigerous cyst and odontogenic keratocyst was given in relation to missing 47 and 48 teeth. A screening orthopantomogram was advised.

The panoramic radiograph revealed impacted 47 and 48 with radiopaque mass surrounded by a radiolucent border measuring approximately 2 cm × 2.5 cm in size distal to 46. The radiopaque mass had displaced 47 anteroinferiorly till the inferior border of the mandible and the 48 posterosuperiorly into the ramus. The radiopaque mass had also displaced the inferior alveolar nerve canal inferiorly [Figure 1].

A radiographic diagnosis of complex odontoma associated with impacted 47 and 48 with superimposed infection was given. A radiographic differential diagnosis of calcifying epithelial odontogenic tumor, the adenomatoid odontogenic tumor was given.

Excisional biopsy along with the extraction of the impacted 47 and 48 was planned under general anesthesia. Under strict aseptic precautions and antibiotic coverage, the tumor was addressed by placing an envelope incision along the ascending border of the ramus extending up to right second premolar. A full thickness mucoperiosteal flap was reflected, and the bone removal was performed with the help of surgical burs. The tumor along with the impacted 47 and 48 was removed carefully without causing damage to the adjacent vital structures [Figure 2]. Since the bony defect was large making the operating site weak, we decided to facilitate early bone regeneration by placing platelet rich fibrin and bio-scaffold mesh shaped to restore the form of the excised portion of the mandible. The patient was administered injection ceftriaxone 1 g intravenously twice daily for 5 days and injection metrogyl 100 ml thrice daily for 3 days which was started a day before surgery.

The tumor mass, impacted tooth with follicle and soft tissue associated with the tumor was sent in three separate labeled containers for histopathological examination. The section comprising of the tumor mass showed a benign tumor composed of strands of cuboidal epithelial cells in two cell layers embedded in a cellular and myxomatous connective tissue stroma [Figure 3]. A dense mixed inflammatory infiltrates comprising of plasma cells, foamy macrophages, neutrophils, lymphocytes, and occasional giant cells were also seen. The section comprising of the impacted tooth with follicle showed tooth structure with no pathology. The section comprising of the soft tissue associated with tumor showed skeletal muscle fibers with extensive granulation tissue formation. Dentinoid

deposition was seen within deeper ectomesenchyme [Figure 4]. A histological impression of infected AFD was given.

The patient was kept under periodic follow-up for next 1½ years which was found to be uneventful [Figure 5]. Patient's mouth opening and function were satisfactory.



Figure 1: Orthopantomogram showing a mixed radiolucent-radiopaque lesion with impacted 47 and 48



Figure 2: Excised specimen

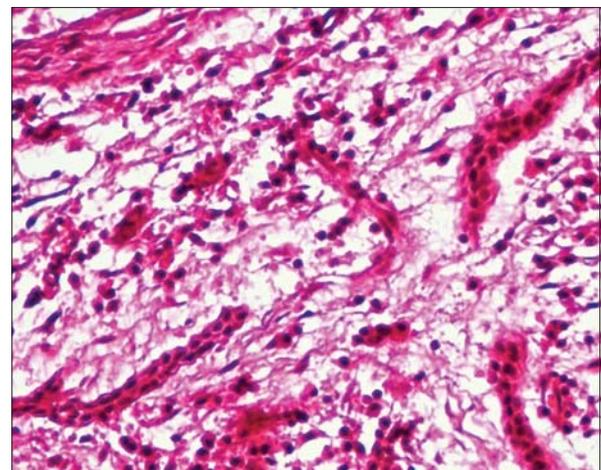


Figure 3: H and E stained section showing strands of cuboidal epithelial cells in two cell layers embedded in cellular and myxomatous connective tissue stroma

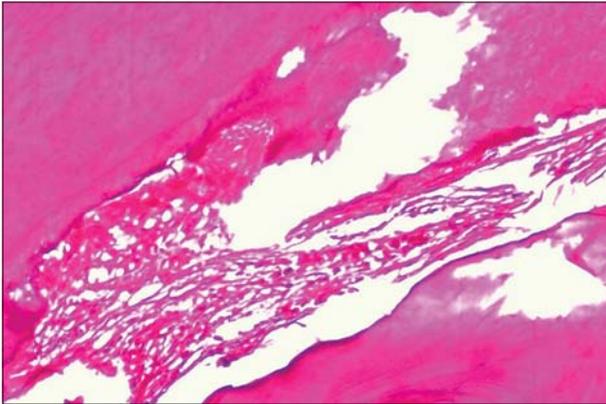


Figure 4: H and E stained section showing dentin formation within deeper ectomesenchyme

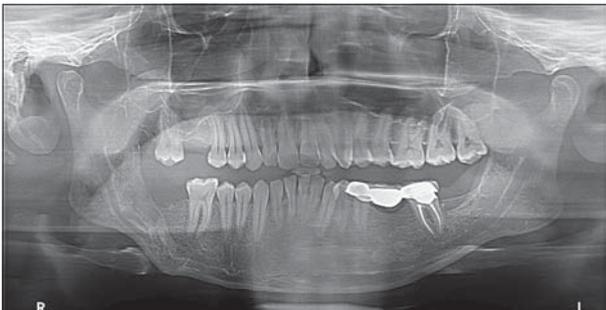


Figure 5: 1½ year post-operative orthopantomogram showing adequate bone formation at surgical site

Discussion

The existing literature concerning the interrelationship of the group of odontogenic lesions consisting of odontogenic epithelium with odontogenic ectomesenchyme with or without dental hard tissue formation also referred as mixed odontogenic tumors is still controversial. These mixed tumors include ameloblastic fibroma, AFD, and ameloblastic fibro-odontoma.^[2] The AFD is one of the rarest, mixed odontogenic neoplasms consisting of neoplastic odontogenic epithelium and mesenchyme with dentin or dentin-like tissue.^[3] Only 34 additional cases have been published up to 2012 since the first case reported by Straith, in 1936, according to PubMed search conducted by Umashankara *et al.*^[4]

AFD is a controversial tumor considering its biological nature and histological diagnosis. It has been suggested that AFD is not a true neoplasm but a hamartoma. The limited growth potential of this tumor may support this suggestion.^[3] However, the WHO publication on histological typing of odontogenic tumors defined AFD as “a very rare neoplasm composed of odontogenic epithelium and an immature odontogenic connective tissue and characterized by the formation of dysplastic dentin.” It has been reported that AFD may have a significant growth potential and may enlarge to a considerable size supporting the statement that this lesion is a true neoplasm.^[3]

Some regard ameloblastic fibroma, AFD, and ameloblastic fibro-odontoma as separate entities while some consider them as chronological stages beginning as ameloblastic fibroma at the outset and progressing into AFD, ameloblastic fibro-odontoma, and odontoma successively. In support of this proposition, Slootweg analyzed 33 mixed odontogenic tumors and found that the mean age of the patients with ameloblastic fibro-odontoma was lesser than that of the patients with ameloblastic fibroma. If his presumption was assumed to be correct, then the patients with ameloblastic fibro-odontoma (which is assumed to differentiate further from ameloblastic fibroma) should have been greater than that of those with ameloblastic fibroma. Therefore, he concluded that ameloblastic fibroma represents a separate neoplastic entity that does not progress into a more differentiated odontogenic lesion.^[5]

The true existence of AFD is not completely accepted. Indeed, AFD is considered, by some authors, to hold a stage between the ameloblastic fibroma and ameloblastic fibro-odontoma based on the extent of histodifferentiation that there may be a maturation spectrum from the ameloblastic fibroma to the ameloblastic fibro-odontoma with the AFD as an intermediate form. However, this does not necessarily suggest that all ameloblastic fibromas will differentiate over time into an ameloblastic fibro-odontoma or an odontoma.^[6,7]

The WHO classification claims that “until more experience has been gained it may be worthwhile separately identifying the differing patterns or types (of ameloblastic fibroma and related lesions), even though some of these may later prove to be nothing more than stage in the evolution of a single type of tumor.” Gardner has an opinion that the AFD should be referred to simply as “ameloblastic fibroma.”^[8]

Philipsen *et al.* suggested that ameloblastic fibroma and AFD manifest in two forms. The first is a neoplastic lesion which if left *in situ* will not mature further. The second variant is a hamartomatous lesion that appears to have the potential to differentiate into an ameloblastic fibro-odontoma and mature further into a complex odontoma. This idea has been incorporated into the suggested modifications for the 1992 edition of the WHO histological typing of odontogenic tumors.^[5]

This tumor arises mainly in the posterior region of the mandible and is usually seen in association with unerupted molar teeth which correlate with our case too. This tumor is more commonly found reported in males with a male to female ratio of 2:1. It is a slow growing often asymptomatic lesion and may enlarge to extreme sizes.^[4]

According to Philipsen *et al.*, AFD was first described by Straith (1936) under the term dentinoma. Subsequently, it was called as immature dentinoma, ameloblastic fibroma with the dentinoid formation and as fibro-ameloblastic type of dentinoma by different authors. Most of the studies showed that AFD occurred in two forms mature and immature dentinoma. Immature form is thought to be an AF in which induction of mesenchymal cells by odontogenic epithelium has resulted in the formation of organic matrix of dentin or osteodentin and in mature type all the elements are similar, but with scanty or absence of an odontogenic epithelium. According

to Takeda, immature dentinoma is different from AFD as both epithelial and fibrous elements may resemble those of odontogenic fibroma rather than that of AF with or without dentin formation.^[9] This tumor originates mainly in the posterior region of the mandible and is usually seen in association with unerupted molar teeth which correlate with our case too. This tumor is more commonly found reported in males with male:female ratio of 2:1. It is a slow growing often asymptomatic lesion and may enlarge to enormous sizes.^[4] AFD radiographically shows a fairly well-delineated radiolucency with varying degrees of radiopacity. Developing odontomas and ameloblastic fibro-odontoma may also present with similar radiographic appearance and distinction may be difficult.^[1]

The histological features of ameloblastic fibroma include odontogenic ectomesenchyme that resembles dental papilla and strands or islands of odontogenic epithelium that resemble dental lamina and enamel organ. If there is dentin formation the lesion should be diagnosed as AFD; if it is also accompanied by enamel formation, it should be diagnosed as ameloblastic fibro-odontoma.^[2] The presence of both dentin and enamel is essential to call the tumor an ameloblastic fibro-odontoma. Exclusive dentin formation in an otherwise identical tumor is called AFD. A conservative approach is usually recommended for this tumor.^[1]

Since the lesion is benign, the recurrence rate is very low. Occasionally ameloblastic fibrodentinoma, a very rare malignant odontogenic neoplasm is thought to arise from the malignant transformation of the ectomesenchymal component of AFD.^[4] The 2005 WHO classification of odontogenic sarcomas presented two tumors: Ameloblastic fibrosarcoma and ameloblastic fibrodentinoma. In the malignant counterpart, only the mesenchymal component undergoes a malignant transformation while the epithelial component does not show any malignant changes. According to review of the literature by Giraddi *et al.*, on ameloblastic fibrodentinoma and ameloblastic fibro-odontosarcoma, only 15 cases have been reported in literature till 2009.^[2] Giraddi *et al.* reported an aggressive atypical AFD presenting with resorption and perforation of cortical plate which was treated radically.^[2]

This paper emphasizes the importance of a systematic diagnostic approach as a key to early diagnosis, management and hence prognosis of the patient. In cases of delayed eruption, missing or impacted teeth one must understand the importance of an early radiographic examination to rule out any pathology. As our patient had experienced symptoms with the tumor previously, a complete workup at an earlier stage would have helped in an earlier diagnosis and management of the patient. This paper also reports a rare case of a mixed odontogenic tumor which has been reported very scarcely in literature.

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