Ameloblastic Fibrodentinoma: A 12 Years follow-up of a Rare Entity

Oomen Aju Jacob, S K Padmakumar, Devu Aloka, S Sooraj

Ameloblastic fibrodentinoma (AFD), according to the WHO is a neoplasm similar to ameloblastic fibroma, but also showing inductive changes that lead to the formation of dentine. AFD is a very rare odontogenic mixed tumor and only <40 cases have been reported so far. The origin, like other mixed odontogenic tumors is from the odontogenic apparatus. Two schools of thought exist, one to consider it as a variant of ameloblastic fibroodontoma and the other categorize it as a separate entity. An AFD in an 1½-year-old male patient with 12-year follow-up after treatment is presented.

Keywords: Ameloblastic, Fibroodontoma, Odontoma

INTRODUCTION

Ameloblastic fibrodentinoma (AFD), according to the WHO is a neoplasm similar to ameloblastic fibroma (AF) but, also showing inductive changes that lead to the formation of dentine. The origin, like other mixed odontogenic tumors is from the odontogenic apparatus. Only <40 cases have been reported so far. Many authors are of the view that it must be considered as a variant of ameloblastic fibroodontoma (AFO). However, the WHO suggested that though all mixed odontogenic tumors are closely related and may be a part of the same hierarchy, they should be considered as separate entities unless proven otherwise. The clinical presentation is usually as a non-tender, slow growing swelling of the jaw. Radiographically, it can present in varied patterns, most commonly as radiolucency with flecks of radiopacities. An AFD in a 1½-year-old male patient with 12-year follow-up after treatment is presented.

CASE REPORT

A 1½-year-old male patient presented to the maxillofacial surgeon in 2001 with a hard, non-ulcerated, non-tender bony swelling of the right mandible, which was a chance finding by a pediatrician on general examination. The patient’s medical history was unremarkable and no regional lymphadenopathy was noted.

Computed tomography (CT) showed a large expansile lesion (4.6 cm × 2.8 cm) extending from the right angle of mandible, anteriorly into the body and posteriorly into the ramus. The coronoid process was involved although the condyle was intact. There was evidence of cortical expansion and calcifications were seen in the intraosseous component of the lesion. Multiple loculations were also seen at the inferior part of the lesion. In the ramus of the mandible, an impacted tooth was seen within the lesion. An impression of ameloblastoma was made from the CT scan (Figure 1).

Incisional biopsy was done elsewhere prior to referral to the maxillofacial surgeon was reported as odontoameloblastoma. The child was posted for surgery and a soft tissue specimen of size 5 cm × 3 cm was enucleated along with multiple teeth like calcifications resembling odontoma.

Figure 1: Computed tomography sections (a and b) showing the large expansile lesion involving mainly the right angle of mandible, associated with impacted tooth, extending anteriorly into the body and posteriorly into the ramus of mandible.
**Histopathology**

Soft tissue along with hard tissue specimens resembling a tooth like structures was received. The hematoxylin and eosin stained sections of the soft tissue specimen showed a highly cellular primitive connective tissue stroma and discrete islands of odontogenic epithelium (Figure 2).

The tumor islands were lined by tall columnar epithelium resembling ameloblasts with central stellate reticulum like cells. Cystic degeneration was present in a few follicles. Attempted matrix formation, resembling dentinoid material, was seen around many of the islands (Figure 3).

The final diagnosis was AFD. The patient has been followed up postoperatively for 12 years. No evidence of recurrence has been noticed till date.

**DISCUSSION**

The term AFD was introduced by Straith in 1936 and he defined it as “a very rare neoplasm composed of odontogenic epithelium and immature connective tissue, and characterized by the formation dysplastic dentine.”

The term mixed odontogenic tumor is used to indicate those lesions with both neoplastic hard tissue and soft tissue components. To this group belongs AF, AFO, AFD and odontoma.

Differentiating these lesions from AFD requires knowledge about the pathogenesis, demographics and the roentgenographic and histologic features of all of them.

AF and related lesions are defined as neoplasms composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles the dental papilla, and with varying degrees of inductive changes and dental hard tissue formation. AF is a mixed tumor with neoplastic epithelial and mesenchymal components. Variants of AF have been reported namely, granular and peripheral. The continuum concept purporting AF as a stage in the development was proposed by Cahn and Blum. However, the evidence for non-progressive AFs and those arising even beyond the common age of occurrence of odontomas are there. The malignant counterpart, ameloblastic fibrosarcoma (ameloblastic sarcoma) has also been described.

Moreover, Philipsen et al. proposed the hypothesis of neoplastic and hamartomatous line of development for AF and related lesions. Neoplastic lines include AF, which occurs beyond the age of 20 and is closely related to AFD. The maturing capacity of AFD has not been proved yet. The hamartomatous line includes AF which might be part of a developing complex odontoma and AFO. However, there is no modality to differentiate between these two kinds of AFs.

AFO is a hamartomatous lesion similar to AF and AFD, but showing further inductive changes that lead to the formation of enamel matrix in addition to dentine (dentinoid). More than 110 cases have been reported so far. Though radiographically difficult to distinguish, AFO can be differentiated histologically from AFD by the presence of enamel.

Odontoma is a hamartoma with frank mature hard tissue production. Initial stages of odontomas are radiographically as well as histologically similar to AF and at times AFO and AFD. Odontomas have a mixed radiographic presentation and progresses to radio-opacity as it matures. The demographic details of all mixed odontogenic tumors are compared in Table 1 (Compiled from Reichart PA, Philipsen HP. Odontogenic tumours and allied lesions. London: Quintessence Publishing Co Ltd; 2004).

AFD is a tumor of younger age and commonly presents as a slow growing, non-tender swelling, usually associated with a tooth. The mandible is more commonly affected. Site wise, AFD is commonly associated with incisors in the...
deciduous dentition and molars in a permanent dentition. AFD has also been reported associated with anodontia of permanent tooth. The usual radiographic presentation is a well-defined radiolucency with flecks of opacities. AFDs have been known to grow large in size with significant growth potential. Aggressive and pigmented variants have also been reported. Surgical excision is the usual treatment of choice. Recurrence is extremely rare though a malignant variety of the ectomesenchymal component of AFD have been reported, namely AFD sarcoma (AFDS). The propensity for the lesion to recur and show malignant tendencies points more towards its neoplastic nature. Malignancies arising from AFO have been termed AFO sarcoma (AFOS). Numerous AFOS/AFDS cases have been reported so far. Multiple surgical interventions on recurring lesion are thought to be an initiating factor for malignant transformation. Presence of dentinoid differentiates AFDS from AFOS though it has no prognostic implications.

In the present case, the lesion was enucleated along with the involved tooth buds. The patient has been followed-up for the past 12 years. Healing was uneventful and no signs of recurrence have been noted till date. Serial orthopantomograms taken during the past 12 years are shown in Figures 4a-i and 5 shows immediate postoperative CT (a and b) and 3-D CT scan (c), taken 12 years later, showing unhindered and completed growth of right posterior body, ramus and coronoid processes of mandible.

**CONCLUSION**

Because clinical behavior and radiographic features are similar to AF and AFO, diagnosing AFD is considered as an academic exercise for the oral pathologist. Though recurrences are rare, malignant transformation potential of AFD cannot be considered trivial. Even though most cases of AFD behave in a non-aggressive manner, a long-term evaluation and follow-up are mandatory.

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Dr. Lovekesh, Junior Resident, Department of Oral Medicine and Radiology, Government Dental College, Thiruvananthapuram.
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