

Ameloblastic Fibrodentinitoma - A Case Report

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Abstract

The Ameloblastic fibrodentinitoma (AFD) is a minor variation of an Ameloblastic fibroodontoma with a predominant dentin formation. It is a rare mixed odontogenic tumour composed of neoplastic odontogenic epithelium and mesenchyme with dentin. This paper presents a rare case of Ameloblastic fibrodentinitoma in the anterior mandible, which was excised and post-operative recovery was uneventful.

Keywords: AFD; Odontogenic Tumour; Mixed Tumour; Ameloblastic Fibroma

Introduction

The Ameloblastic fibrodentinitoma (AFD) is a rare benign tumor, belong to mixed odontogenic tumour. There is a limited proliferation of odontogenic epithelium of the enamel organ and odontogenic mesenchyme of the primitive dental pulp. This results in soft tissue recapitulation of the dental follicle, which produces predominantly dentin. This group of lesion includes Ameloblastic fibroma (AF), AFD, Ameloblastic fibroodontoma (AFO). The extent of histodifferentiation of AFD is considered to be intermediate stage between the AF and AFO [1]. It is a rare occurrence with less than 1% of odontogenic tumor and more in males compared to females in the ratio of 3:1 [2].

According to WHO Ameloblastic fibrodentinitoma (AFD) is not a true neoplasm and has features similar to ameloblastic fibroma. In addition there are inductive changes that lead to the formation of dentin [3]. AFD is also called as Dentinoma [4]. It presents as a slow growing, asymptomatic jaw expansion. It presents as intraosseous as well as extra osseous lesions. It is not an invasive proliferation, which may attain quite large size and cease to grow. It may resorb the tooth roots and displace developing teeth. The common site for an occurrence is posterior mandible. It occurs mostly in children's and young adults. Radiographically presents as mixed radiolucent radiopaque lesion with a clear demarcation from the adjacent bone. The calcifications are denser and are similar to density of teeth.

Case Report

A 40 year old patient reported with a chief complain of swelling in the lower jaw since 11 years. Patient gives history of increasing swelling in lower jaw which was peanut in size. Patient's medical history was found to be non-significant. On examination a 3 × 3 cm swelling was present in the symphysis region extending from 33 to 43 on the labial side. It was sessile, mucosa was normal with no secondary changes. On palpation the swelling was bony hard and non-tender. Teeth in the region were vital. Pre operatively differential diagnosis of CEOT, mixed odontogenic tumour was given.



Figure 1: Intra oral picture of the lesion.

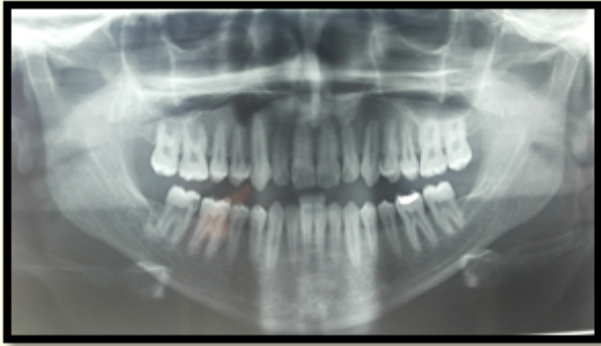


Figure 2: Orthopantomogram showing mixed lesion in anterior mandible

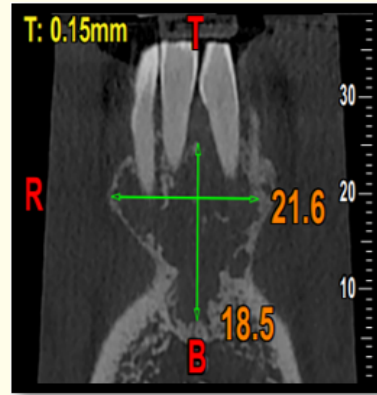


Figure 4: CBCT showing superoinferior and mesiolateral extension of the lesion.

OPG showed an ill-defined radiolucency with scattered foci of calcification with a sclerotic margin. CBCT showed an expansile lesion in lower anterior region extending from 33 to 43, with calcifications inside the lesion. Aspiration showed to be solid tumour. Incisional biopsy reported a benign odontogenic tumour. Under LA full thickness mucoperiosteal flap was elevated from premolar to premolar region, lesion was excised, 31 and 41 teeth were extracted, primary closure was done. Histopathological examination revealed strands and islands of odontogenic epithelium with rich primitive ectomesenchyme. The islands were made of tall columnar cells with reverse polarity and cluster of island showed stellate reticular like cells. Perifollicular dentinoid like material was seen. Over all features were suggestive of ameloblastic fibrodentinoma. 2 years follow up showed no recurrence.

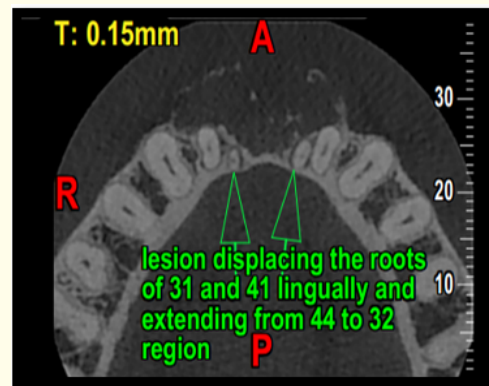


Figure 5: CBCT showing buccal expansion of the lesion.

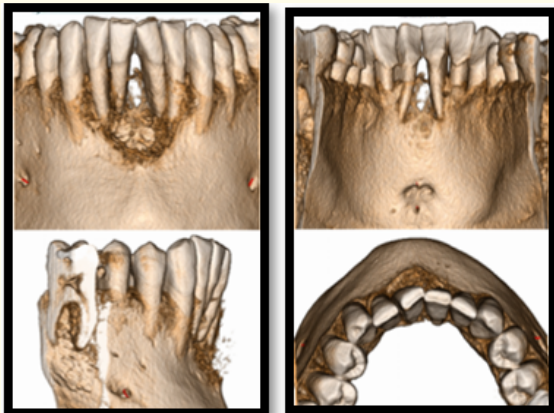


Figure 3: CBCT view showing the bony defect.



Figure 6: Excised specimen along with 31 and 41.

Discussion

Odontogenic tumours arise from the Odontogenic epithelium, ectomesenchyme and mesenchyme. Mixed Odontogenic tumours are a heterogeneous group of lesion with diverse clinical and histopathological features. Ameloblastic fibroma, Ameloblastic fibroodontoma, Ameloblastic fibrodentinitoma and Odontoma belong to this group. The inter relationship between odontogenic tumours, regarding origin histodifferentiation and maturation is often debated. Ameloblastic fibroma is composed of proliferating odontogenic epithelium and mesenchymal components. The varying degree of inductive changes occurs in the mesenchymal components leading to formation of enamel and dentin [5]. In Ameloblastic fibroodontoma there are mesenchymal inductive changes that lead to formation of hard tissues like enamel and dentin [8]. In Ameloblastic fibrodentinitoma there is predominant dysplastic dentin formation [6].

Radiographically it appears as a mixed radiolucent - radiopaque lesion. Various differential diagnosis to consider are ameloblastic fibrodentinitoma, calcifying epithelial odontogenic tumor, calcifying odontogenic cyst and adenomatoid odontogenic tumor. Ameloblastic fibro-dentinitoma is a controversial neoplasm with respect to its biologic nature and histological diagnosis - it is suggestive that Ameloblastic fibrodentinitoma is not a true neoplasm but a hamartoma. The exact histo-genesis of Ameloblastic fibrodentinitoma is unclear. It is thought to fall in a stage between the Ameloblastic fibroma and Ameloblastic fibroodontoma in histo-genesis. Straith first introduced the term Ameloblastic fibrodentinitoma in 1936 and defined it as "a very rare neoplasm composed of odontogenic epithelium and immature connective tissue and characterised by the formation of dysplastic dentin [7,8]. Differentiating Ameloblastic fibrodentinitoma from other mixed odontogenic tumour like Ameloblastic fibroma, Ameloblastic fibroma odontoma and odontoma requires knowledge about the pathogenesis, radiographic appearance and histological features of all of them. Ameloblastic fibrodentinitoma is the commonly seen in younger age group, with most patients being below 30 years. The most commonly involved site is posterior mandible as compared to maxilla. However, in the present case the age of the patient was 40 years and site involved was anterior mandible rarity. According to Philipsen., *et al.* AF and AFD occurs in two variants as non-neoplastic, hamartomatous variant in which lesion is histologically differentiated as AFD and may mature further into complex odontoma. Another variant is neoplastic variant which does not mature further [9].

Rarely the mesenchymal component of Ameloblastic fibrodentinitoma undergoes malignant transformation leading to a fibro-dentinito sarcoma. Hence if the lesion presents with an aggressive behaviour, a radical excision should be performed. Ameloblastic fibrodentinitoma more commonly presents as a benign lesion with a less likely recurrence once enucleated or excised.

Conclusion

Ameloblastic fibrodentinitoma (AFD) is a rare benign mixed odontogenic tumour which is non-invasive. This is found more in children and young adults. Ameloblastic fibrodentinitoma should be considered in the differential diagnosis of mixed lesions. The treatment of choice is surgical excision and the recurrence is rare. Hence a thorough clinical, radiological and histological evaluation with long term follow up is necessary.

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