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Extrafollicular Adenomatoid Odontogenic Tumor: An Unusual Presentation

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ABSTRACT

Adenomatoid odontogenic tumors (AOT) are uncommon, nonaggressive tumors of odontogenic epithelium and have a variety of patterns mixed with mature connective tissue stroma. AOT is frequently asymptomatic and is revealed during a routine radiographic examination or when radiographs are made to determine why a tooth has not erupted. This report describes morphological characteristics, clinical course and surgical treatment of an adenomatoid odontogenic tumor that developed in the right mandible of a 13 year old patient. The patient was treated with surgical excision with no signs of any recurrence

Keywords: Adenomatoid odontogenic tumors; jaw tumors, benign

INTRODUCTION

Adenomatoid odontogenic tumors (AOT) are uncommon, nonaggressive tumors of odontogenic epithelium and have a variety of patterns mixed with mature connective tissue stroma. The term "adenomatoid odontogenic tumor" proposed by Philipsen et al.[1] indicates that it was not a variant of ameloblastoma. In the World Health Organization classification of odontogenic tumors established in 1971, AOT was mentioned as a mixed odontogenic neoplasm.[2]

There are 3 variants of adenomatoid odontogenic tumor, the follicular type (accounting for 73% of cases), which has a central lesion associated with an embedded tooth; the extrafollicular type (24% of case), which has a central lesion and no connection with the tooth; and the peripheral variety (3% of cases).[3,4] AOT is frequently asymptomatic and is revealed during a routine radiographic examination or when radiographs are made to determine why a tooth has not erupted [5]. A delayed eruption of a permanent tooth or a swelling of the jaws may be the first symptom[6]. Larger lesions cause painless expansion of the bone.[7]

The WHO histological typing of odontogenic tumors, jaw cyst and allied lesions (2005) has defined AOT as a tumor of odontogenic epithelium with duct-like structures and with varying degree of

inductive changes in the connective tissue.[8] Conservative surgical enucleation is the most suggested choice of treatment. Recurrence rate for AOT is exceptionally rare.

CASE REPORT

A 13-years-old male patient reported to the Department of Oral medicine and Radiology with a complaint of swelling on his lower right front tooth region since 6 months. History of the present illness revealed that initially the swelling was small in size and gradually it increased to reach up to the present size. It was not associated with any pain or discharge with no history of trauma associated with it. No aggravating and relieving factors were reported. He had difficulty during tooth brushing, chewing food. Extra oral examination revealed swelling over the upper lip on the right side (Figure 1a). Intraoral examination (Figure 1b and 1c) revealed an exophytic growth of gingiva measuring about 2 cm x 2 cm in size. It appeared pale in a few areas and erythematous in certain area colour, in relation to the mandibular right canine-premolar region, extending from distal to the right lateral incisor to the mesial to the right premolar and up to the level of occlusal surface. On palpation, the swelling was firm in consistency, nontender with sessile base. In hard tissue examination, all teeth were present. Mandibular right canine was mesially displaced and mandibular right lateral incisor was tilted lingually. Calculus and stains were minimal. So, based on the history and clinical examination a provisional diagnosis of reactive or inflammatory fibrous hyperplasia was given with differential diagnosis of giant cell granuloma, peripheral ossifying fibroma, dentigerous cyst, adenomatoid odontogenic tumor, peripheral ameloblastoma, adenomatoid odontogenic tumor.

In investigations OPG, IOPA-radiograph and cross-sectional mandibular occlusal radiograph, complete blood test, bleeding time, clotting time, random blood sugar, haemoglobin level, complete blood count, differential count, hepatitis and HIV test, serum calcium and paratharmone levels and excisional biopsy were done.

Intraoral periapical radiograph (Figure 1d) showed mixed lesion, in which sclerotic radiodense mass in between mandibular right canine and mandibular right lateral incisor was seen. Tilting of the mandibular right lateral incisor resulted in the overlapping of the crown. Intraoral periapical radiograph (Figure 2a) showed mesial displacement of canine and a well-defined solitary radiodense mass is seen in between mandibular right first and second premolars.

Occlusal radiograph shows (Figure 2b) the lesion is a well-defined corticated border with alteration of the normal contour in buccal and lingual surface, suggested a expansion in buccal and lingual surface.

Panoromic radiograph shows (Figure 2c) a well-defined round unilocular radiolucent lesion is seen in right parasymphyseal region. The lesion is extending from the distal surface of mandibular right lateral incisor distally till the mesial surface of mandibular right second premolar. Superior extension of the lesion is not appreciated. The lesion extends inferiorly beyond half the mandible. The lesion has an uniform well defined corticated border. Distal tilting of mandibular right lateral incisor was noted and mandibular right canine was mesially displaced. Pulpal calcification was present in all first molars. Soft tissue shadow of the swelling is observed on the occlusal surfaces between mandibular right canine and mandibular right first premolar.

Complete blood investigation of the patient was carried out and it was found to be normal. Surgical excision was carried out and the specimen was sent for the histopathological examination which revealed cuboidal to columnar cells arranged in form of nests and rosettes. Some areas showed arrangement of cells in the form of different sized whorls with evidence of ductal pattern. Convoluted structures were seen at the periphery with few cells

arranged in form of sheets as well. Areas of fibrous connective tissue showing presence of fibroblast, blood vessels and extravasated RBCs is also noted, which confirmed the final diagnosis of adenomatoid odontogenic tumor (Figure 2d).

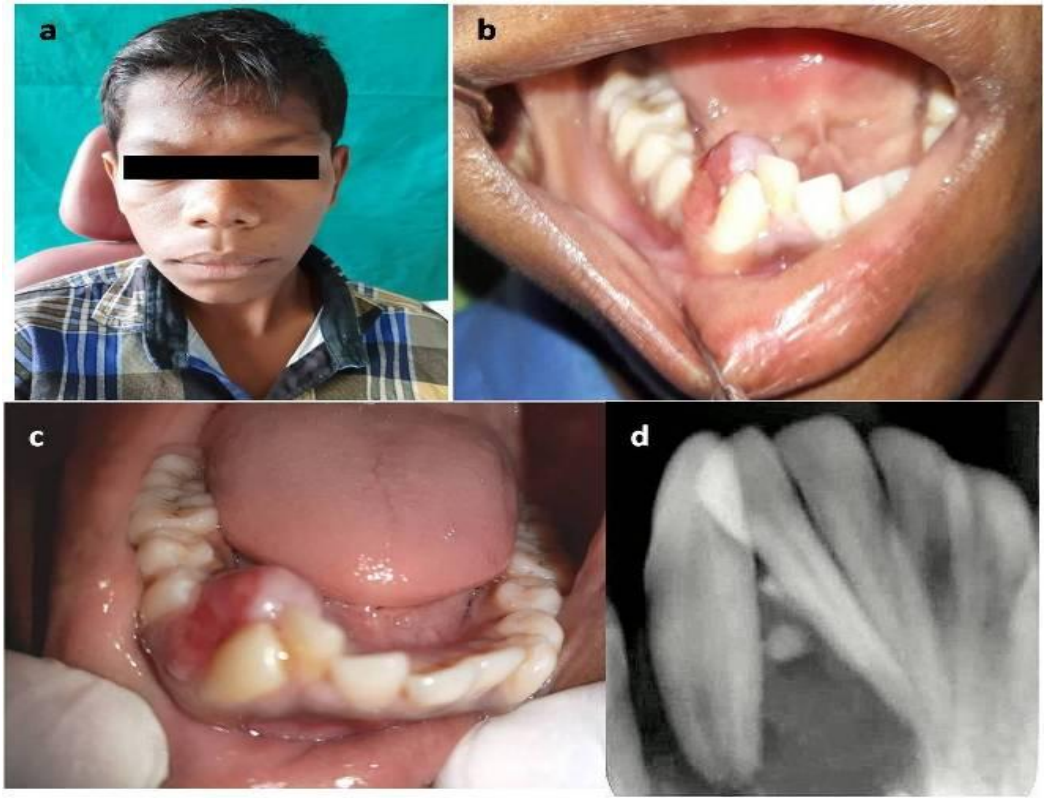

Figure 1. a, b, c- Clinical examination; d-IOPA radiograph

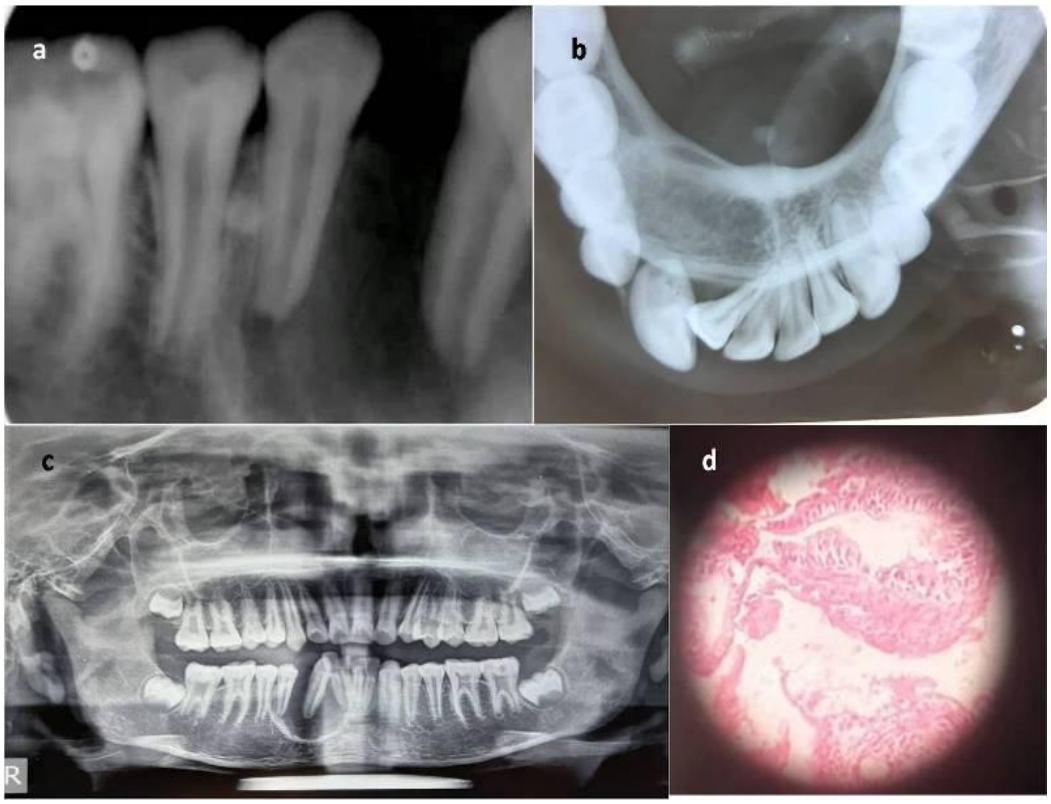

Figure 2. a-IOPA radiograph; b-occlusal radiograph; c-OPG; d-histological section

DISCUSSION

AOT is a benign, non-invasive odontogenic lesion showing slow growth. It is generally intraosseous, but can also occur rarely in peripheral locations.[8] Jörg GK Handschel et al(2005)-From the early 1990s onwards 65 single cases of AOT have been published.[9] The mean age was 13.2 years (range 3 until 28Years) The female: male ratio was 2.3 : 1.[10]

The AOT was predominantly found in the upper jaw. There is 2:1 female to male ratio for all age groups and all variants. The size of the lesion ranges from 2 to 7 cm with a slow growing pattern which results in a painless expansion of the jaws. [10]

AOT is most frequently associated with a missing permanent tooth (maxillary canine to be most frequent). Adjacent teeth are displaced by the growing tumor. This feature is seen in the presented case.

Here, the presented case is extrafollicular type. The radiographic features of AOT many times resemble other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumor, ameloblastoma, odontogenic keratocysts and periapical diseases.[11] Apart from classical unilocular well-circumscribed radiolucency, AOT also shows some small radiopaque foci indicative of small calcifications in tumor mass. The flacks of radio opacity were better appreciated with an intraoral periapical of the affected region. Approximately 78% of AOT shows those calcified deposits.[12] In this case sclerotic radiodense mass in between 42 and 43 was seen. Tilting of the 42 result in the overlapping of the crown. Intraoral periapical radiograph also shows mesial displacement of canine and a well-defined solitary radiodense mass is seen in between 44 and 45.

The origin of this tumor is controversial. The dental lamina remnants are likely to represent the progenitor cells as it not only arises from anterior maxilla but also, as seen in this case, in the body of mandible. In this case, the lesion surrounds a permanent mandibular first premolar and second premolar. The lesion is well-capsulated and so once excised, the recurrence is very rare. Only three cases of recurrence have been reported in Japanese patients.[10]

All the variants of AOT show identical histology. WHO defines AOT as a tumor of odontogenic epithelium with duct like structures and with varying degrees of inductive changes in the connective tissue. The tumor may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst.[13] In this case, cuboidal to columnar cells arranged in form of nests and rosettes were observed. Some areas showed arrangement of cells in the form of different sized whorls with evidence of ductal pattern. Convoluted structures were seen at the periphery with few cells arranged in form of sheets as well. Areas of fibrous connective tissue showing presence of fibroblast, blood vessels and extravasated RBCs is also noted.

Treatment is usually conservative, but follow-up is necessary. The patient was treated with surgical excision under general anesthesia and then was followed up for 6 months. During this phase, the defect left after the treatment resolved well and no signs of recurrence were seen.

CONCLUSION

Ademato id odontogenic tumor (AOT) is generally asymptomatic but sometimes it may cause cortical expansion displacing the adjacent teeth. The growth of the tumor results in facial deformities. Early diagnosis with excisional surgical treatment helps in the prevention of extensive bone destruction.

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