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A Case Report: Adenomatoid Odontogenic Tumor with Impacted Premolar in Maxilla

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Abstract **Case Report**

Adenomatoid odontogenic tumor (AOT) originates from odontogenic epithelium. It accounts for 3-7% of all odontogenic tumors. It is a benign (hamartomatous), non-invasive lesion with slow progressive growth. AOT is often misdiagnosed as an odontogenic cyst. It is predominantly found in young female patients, and more often in the maxilla than mandible. Frequently associated with an unerupted permanent tooth. AOT resembles dentigerous cyst and tumors like ameloblastoma. These lesions are managed conservatively by surgical excision followed with the removal of the affected tooth and reported to have a good prognosis. Here with, we report a case of AOT in maxillary anterior region in 18-year-old female patient treated conservatively and with good prognosis.

Keywords: Hamartomatous, Odontogenic tumor.

INTRODUCTION

Adenomatoid odontogenic tumor(AOT), is an uncommon benign epithelial lesion. It was first "described by Drieibaldt in 1907[1]." It is odontogenic in origin. The name adenomatoid odontogenic tumour was given by Philipsen and Birn in 1969 and they suggested that it should not be regarded as a variant of ameloblastoma because of its varying behaviour [2, 3]. According to World Health Organization (WHO) "Histological typing of odontogenic tumors[4,5]," AOT is defined as "A tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change 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."

AOT is also called 'two-thirds tumor,' because 2/3rd occur in young females, 2/3rd of tumors occur in the maxilla, 2/3rd are associated with un-erupted teeth, and two-thirds of the affected teeth are canines[5]. The variants of AOT are follicular variant (73%), it is a central lesion associated with an embedded tooth, the extrafollicular variant (24%), which has a central lesion and no connection with the tooth and the peripheral variant (3%)[6,7]. The tumor causes drifting of adjacent teeth as tumor expansion is more common than tooth root resorption. Radiographically, it presents as a unilocular mass involving an unerupted tooth, opaque in the center and sclerotic at the periphery. Conservative management through surgical enucleation is the widely preffered treatment of choice, while recurrence rate is exceptionally rare [4].

CASE REPORT

An 18-year-old female patient reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of swelling in the right maxillary region since 8 months. Extraorally, a diffuse swelling of approximately 4×3cm was observed over the right middle third of the face. The swelling extended superiorly from ala tragus line; inferiorly to the corner of mouth; anteriorly ala of the nose; posteriorly 6cm ahead of the tragus.

Intraoral examination revealed an expansile swelling extending from right maxillary canine to first molar on the same side with obliteration of buccal vestibule and missing maxillary right first premolar. Mucosa overlying the swelling was normal .On palpation swelling was tender. On aspiration, blood tinged serous fluid was obtained.

OPG and occlusal cross-sectional radiographs were taken, which showed a well-defined unilocular radiolucency associated with impacted first premolar (Fig1). Based on clinical and radiological findings, the

differential diagnosis of dentigerous cyst, unicystic ameloblastoma, odontogenic keratocyst and AOT were made.

The mass was enucleated completely along with the embedded 1st premolar and distal tilted 2nd premolar was also extracted, under local anesthesia (Fig2) and sent for histopathological examination. Gross examination of the specimen exhibited a mass of 4×3 cm in size which was adhered to the root apex of impacted tooth (Fig 3).

Microscopically, nodules of varying sizes of cuboidal or columnar epithelial cells that formed nests, whorls, and rosette like structures are seen. Foci of eosinophilic hyaline droplet material and calcifications were also observed. The stroma of the connectivetissue was loosely arranged and contained thin-walled congested vessels. Duct like structures were located in many of epithelial nodules. The overall histopathological features were suggestive of AOT.

Post-operative radiographs were taken for documentation including OPG & Occlusal view of maxilla. Post-op OPG showed enucleated lesion along with extracted impacted maxillary 1st premolar and 2nd premolar. Occlusal view revealed decreased size of cortical expansion. Repeated follow-ups were advised both clinically and radiographically, we observed gradual reduction in the size of the deformity and adequate healing of the defect (Fig:4).

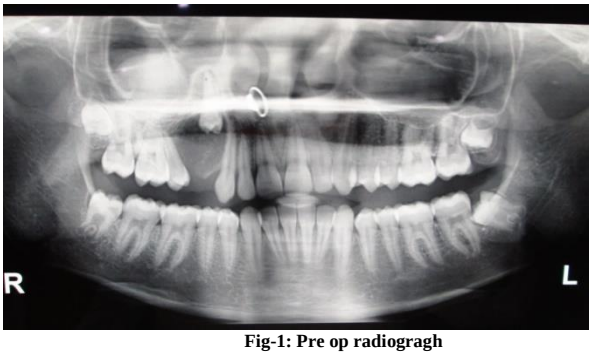

Fig-1: Pre op radiogragh

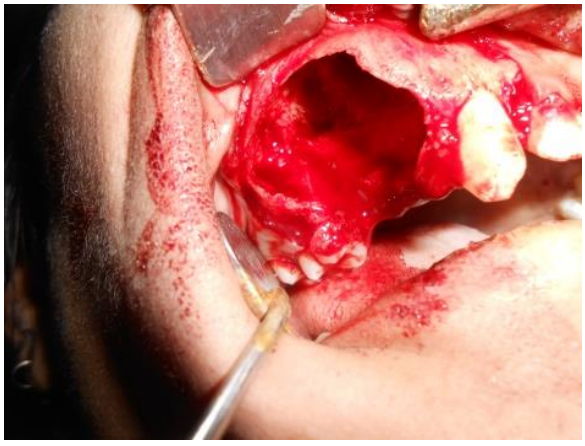

Fig-2: Intra op photograph

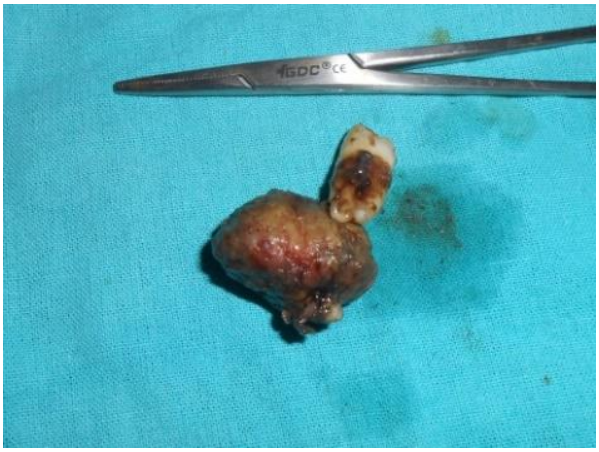

Fig-3: Enucleated mass

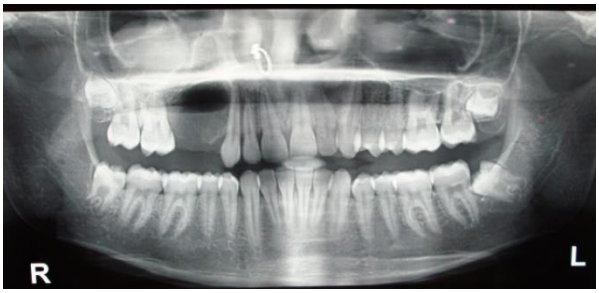

Fig-4: Post op Radiograph

DISCUSSION

AOT is a benign, non-invasive slow growing odontogenic tumour. It presents as intraosseous in nature, but can rarely occur in peripheral locations. AOT is mostly seen in young patients, in the second decade of life and is uncommon in patients who have crossed 30 years of age. Females are more affected by AOT, with a female to male ratio of 1.9:1[8, 9]. This female predilection is more marked in Asian populations, the highest female incidence being observed in Sri Lanka (3.2:1) and Japan (3:1)[10]. The maxilla is predominant site of occurrence, being almost twice as frequent as that in the mandible, and the anterior part of the jaw is more frequently involved. An unerupted maxillary canine is the tooth most commonly associated with AOT. The case we reported is similar to the reported literature as a female young patient but with impacted maxillary first premolar rather than frequently reported cases of canine impaction. Irregular root resorption is seldom reported. Clinically, AOT presents as a slow-growing symptom free lesion, with central lesion which results in cortical expansion, adjacent teeth may be slightly displaced[11].These features are evident in the described present case.

The origin of AOT is controversial. However, most authors accept its odontogenic source: it occurs within the tooth bearing areas of the jaws and is often found in close association with embedded teeth, having cytological features similar to those of various components of the enamel origin, dental lamina, reduced enamel epithelium and/or their remnants [7, 12]. Some support the idea that the lesion is a

developmental outgrowth or hamartoma while others consider it to be a neoplastic growth of odontogenic epithelium. The 1971 WHO classification stated:" it is generally believed that the lesion is not a neoplasm".

Adenomatoid odontogenic tumor can occur both intraosseously and extraosseously. Intraosseous AOT are of 2 types: follicular (or pericoronal) and extrafollicular (or extracoronal) based on radiographs. The former is interpreted as well defined unilocular radiolucent lesion surrounding the crown and is often part of the root of an unerupted tooth. The latter is a well-defined radiolucent lesion, but located between /above / superimposed upon the root of an unerupted tooth [13]. Minute, variable-shaped radiopacities are frequently found within the lesion. In our case, radiopacities were discernable. The extraosseous, peripheral or gingival types of AOT are rarely detected radiographically, but there may be slight erosion of the underlying alveolar bone cortex [14].

AOT is usually surrounded by a well-developed connective tissue capsule. It may present as a solid mass, a single large cystic space or as numerous small cystic spaces. The tumor is composed of spindle shaped or polygonal cells forming sheets and whorled masses in a scant connective tissue stroma.

As all the variants of AOT reveal an entirely benign biologic behaviour and are well encapsulated, conservative surgical enucleation or curettage has been proven to be the treatment of choice. In the present case, no recurrence wasobserved after one year of follow up.

CONCLUSION

AOT is an uncommon odontogenic lesion, but can be identified from its clinical and radiographic appearance. Persistence of deciduous teeth for a longer duration and unerupted succeeding permanent teeth, when associated with a swelling, always need to be investigated for associated odontogenic lesions.

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