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ADENOMATOID ODONTOGENIC TUMOR IN A 7 YEAR OLD CHILD: A RARE CASE REPORT

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ABSTRACT
Adenomatoidodontogenic tumor (AOT) is a benign odontogenic tumor which constitutes about 2-3% of all odontogenic tumors. It is a hamartomatous, non-invasive leison commonly seen in second decade with female predilection. In most of the cases, it is located more often in the maxilla associated with an unerupted permanent tooth. It is considered as hamartoma because of its limited size and growth potential. The general age incidence of AOT is second decade, but here we report a case of follicular AOT in a 7 year old child which is uncommon.

INTRODUCTION

Adenomatoidodontogenic tumor (AOT) is a distinct odontogenic tumor [1], which was first described by Dreibladt in 1907 as pseudo adenoameloblastoma [2]. The name adenomatoidodontogenictumour was proposed by Philipsen and Birn in1969 [3] and suggested that it not be regarded as a variant of ameloblastoma because of its different behaviour[1]. Adenomatoidodontogenic tumor is also called 'two-thirds tumor,' because 2/3rd of the leisons occur in young females, 2/3rd of tumors occur in the maxilla, 2/3rd of the cases are associated with un-erupted teeth, and two-thirds of the affected teeth are canines [4].

There are 3 variants of adenomatoid odontogenic tumour, the follicular type (70%), extra-follicular type (25%) and peripheral (5%) variant[2]. The histologic typing of WHO (2005) has defined AOT as a tumor composed of odontogenic epithelium presenting a variety of histoarchitectural patterns, embedded in mature connective tissue stroma, and characterized by slow but progressive growth [5]. Conservative surgical enucleation is the choice of treatment. Recurrence rate of AOT is rare [6]. We, here by report a case of AOT arising from permanent maxillary canine in a 7 year old child.

CASE-REPORT

A 7-year-old female child reported to the Department of oral medicine with a complaint of swelling in the right upper front tooth region since 1 month. History of the present illness revealed that initially the swelling was small in size and gradually increased to the present size. It was not associated with any pain or discharge, with no history of trauma associated with it. Extra oral examination revealed mild facial asymmetry (Figure 1). Intraoral examination revealed a solitary diffuse swelling on the right anterior maxillary teeth region extending from mesial aspect of deciduous maxillary canine to mesial aspect of deciduous maxillary first molar obliterating the sulcus which is roughly oval in shape measuring about 3x4 cm. The colour of overlying mucosa was normal. On palpation, all inspectory findings were confirmed. The swelling was soft in consistency and non tender. Based on the history and clinical examination, a provisional diagnosis ossifying fibroma was given. Radiographic examination of orthopantomograph (fig 3) and lateral view of skull (fig 4) showed a well-defined unilocular radiolucency with respect to deciduous maxillary canine associated with impacted permanent maxillary canine.

Root resorption in relation to deciduous maxillary canine was noted (Figure 3). Based on radiographic features, differential diagnosis of adenomatoidodontogenic tumor and dentigerous cyst was given. Incisional biopsy was done and specimen was sent to our department.

Histopathological examination revealed odontogenic epithelial cells arranged in different morphological patterns such as sheets, ducts, whorls and rosettes (fig 5). At high magnification, sheets and nests of polyhedral cells along with ductal pattern are lined by cuboidal to coloumnar cells with hyperchromaticnuclei (fig 6). The presence of rosettes with eosinophilic material filling the ductal lumen along with areas of calcification suggested the diagnosis of AOT. Healing was uneventful with no recurrence after surgery.

Fig 1. A single diffuse swelling is seen on the middle third of face

Fig 2. Swelling seen in relation to deciduous maxillary canine obliterating the vestibule

Fig 3. Lateralcephalogram showing unilocular pear shaped radiolucency in association with permanent maxillary canine.

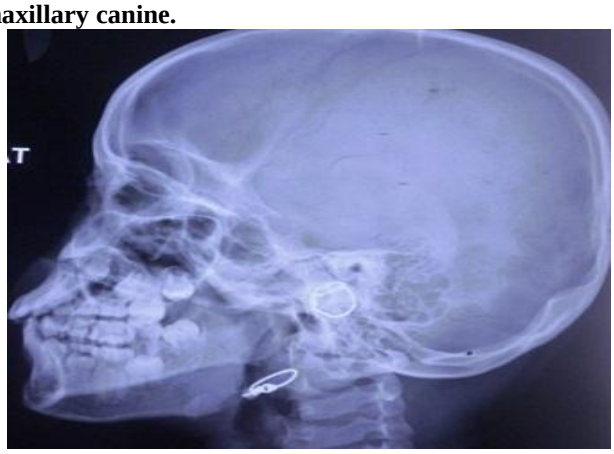


Fig 4. OPG showing unilocular radiolucency in association with permanent maxillary canine.

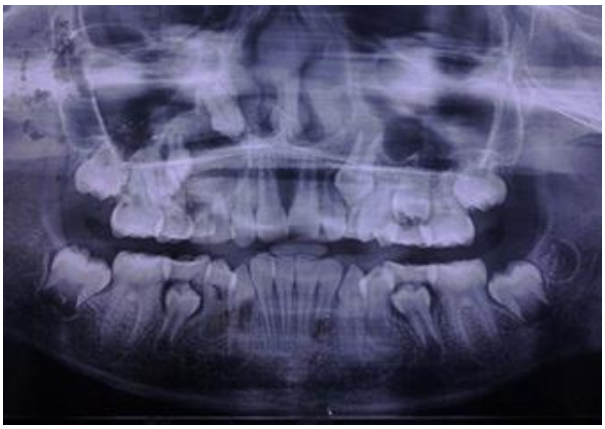


Fig 5. H&E section showing cells arranged in sheets ,ducts and whorl pattern (10x view)

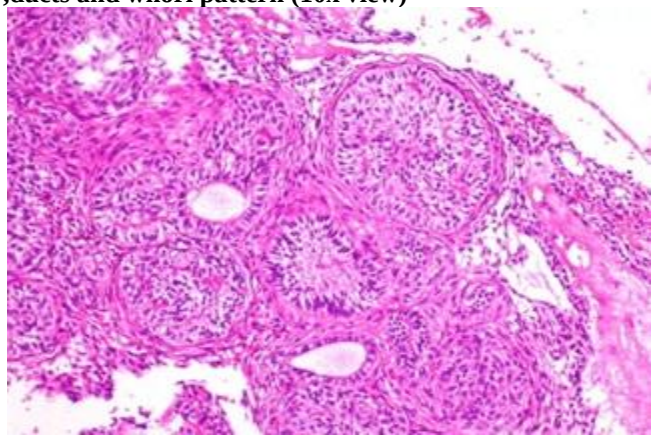
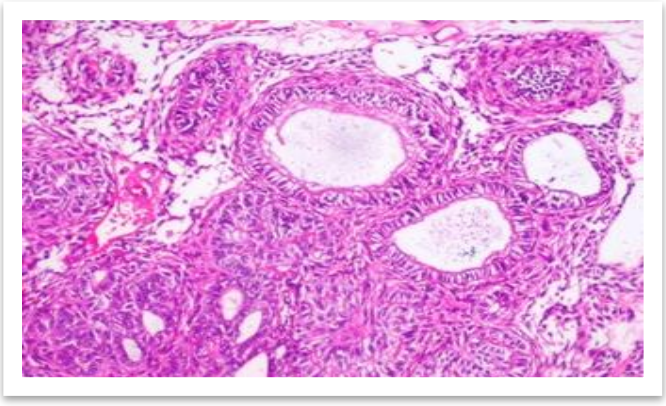


Fig 6. Microscopic photograph showing duct-like structures of odontogenic epithelium lined by a single row of cuboidal or low columnar epithelial cells (H&E ×10).



DISUSSION

Adenomatoidodontogenic tumor is a slow growing leison which comprises 2-3% of all odontogenic tumors [7]. AOT is defined as a tumor of odontogenic epithelium with duct-like structures and varying inductive changes in the connective tissue [8]. Stafne in 1948 recognized it as a distinct clinicopathological entity. The original description of the lesion was given by Gosh in 1934 who named this lesion as an 'Adamantinoma of the upper jaw' [9]. The tumors are usually 1.5 to 3 cms but larger lesions have also been reported in the literature.

The lesions are typically asymptomatic, but may cause cortical expansion and displacement of the adjacent teeth [10]. AOT can occur both intraosseously and extraosseously. Radiographically, intraosseous AOT can be divided into 2 types: follicular and extrafollicular. Intraosseous AOT are more frequent than the extraosseous variant with maximum cases showing the follicular variant. Radiographically, the tumor usually appears unilocular, but cases with multilocular appearance have been reported [11]. The tumor is commonly associated with an unerupted tooth simulating a dentigerous cyst. It may often appear complete radiolucent, however, they contain fine calcifications, a feature that may be helpful in differentiating AOT from dentigerous cyst. The radiolucency associated with an AOT may extend more apically than that of a dentigerous cyst. The most frequent histological pattern is the proliferation of sheets, nests and cords of ameloblast-like cells supported by a scanty often hemorrhagic stroma. These cells may be organized to formwhorls, rosettes, nodules or surround ovoid spaces to form duct-like structures. Additionally, "amyloid" like amorphous eosinophilic material may be found filling or lining the ductal lumens. 78% of AOT shows those calcified deposits.

AOT occurs more commonly in second decade with female predilection but the case we are reporting here is present in a 7 year old child associated with unerupted `permanent maxillary canine which is very rare to occur in that age group. In our case, the tumor was follicular intraosseous type, and was found in the anterior region of the mandible. Radiographic and histopathological findings were suggestive of AOT.

Conservative surgical enucleation is the treatment of choice. Giansanti et al reported that AOT was a completely benign tumor which never recurred once removed. Guided tissue regeneration with membrane technique is suggested for periodontal intrabony defects caused by AOT after complete removal of the tumor.

CONCLUSION

AOT is an uncommon odontogenic lesion which 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 odontogenic lesions. Care should be taken to distinguish such lesion from more common lesions of odontogenic origin during routine dental examinations to prevent extensive surgery.

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