Case Report

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Adenomatoid Odontogenic Tumor: A Case Report

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Adenomatoid odontogenic tumor (AOT) is a very rare odontogenic tumor with an incidence of 1%. Overall it accounts for 9% of all odontogenic tumors. In most of the cases, AOT is misdiagnosed as an odontogenic cyst. Younger individuals are commonly affected and particularly in females. AOT is seen predominantly in the maxillary anterior region in association with an unerupted tooth. Permanent dentition is affected more than the deciduous dentition. Intraoral periapical radiographs play a major role in the diagnosis compared to orthopantomogram because of its increased contrast but for the better assessment of the extension of larger lesions orthopantomogram is must. AOT resembles benign odontogenic lesions like dentigerous cyst and tumors like ameloblastoma. The lesions are managed conservatively by surgical excision along with the removal of the affected tooth and have an excellent prognosis.With this background, we report an unusual case of AOT involving maxillary anterior region in 15-year-old male patient. The present article reviews the etiology, clinical features, histopathological features, and treatment modalities of AOT.

Keywords: Adenomatoid odontogenic tumor, Anterior maxilla, Benign neoplasm

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is an uncommon benign epithelial lesion of odontogenic origin. Previously it was known as pseudo adenoameloblastoma by Dreibladt (1907).[1] Later Staphne (1948) first recognized this as a distinct pathological entity. It constitutes about 2-3% of all odontogenic tumors.[2] Philipsen *et al*. divided AOT as three variants the follicular type; the extra follicular type and the peripheral variety.[3] These variants have common histologic characteristics, which indicate a common origin, this being derived from the complex system of dental lamina or its remnants. Majority of cases constituting of about 88% are diagnosed in 2nd and 3rd decade of life. This tumor is seen in female to male ratio at the rate of 2:1 and has a predilection for the anterior maxilla.[4] Conservative surgical enucleation is the most suggested choice of treatment. Recurrence rate for AOT is rare.[5] Hence, successive unerupted permanent teeth or persistence of deciduous teeth for a longer duration when associated with a swelling should always be suspected for odontogenic lesions.

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CASE REPORT

A 15 years adolescent male child reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling in the upper left front tooth region since 3 months. Initially, swelling was small in size and gradually grew to present size. Swelling was asymptomatic and no history of trauma associated with it.

No relevant medical or family history were contributory. On examination, all vital signs were within normal limits and extraoral examination revealed mild facial asymmetry with the obliteration of the nasolabial fold on left middle 3rd of the face. Surface over the swelling was normal. Swelling was hard in consistency and non-tender on palpation. Ipsilateral left submandibular lymph node was mobile, firm, and non-tender on palpation (Figure 1).

Intraoral examination revealed a solitary diffuse swelling on the anterior maxillary teeth region extending from mesial aspect of 12 to distal aspect of 23, roughly oval in shape measuring about 1 cm × 2 cm in greatest dimension. The color of overlying mucosa was normal. On palpation, all inspectory findings were confirmed and swelling was firm in consistency and non-tender.

On hard tissue examination, there was clinically missing 21 and 22 (Figures 2 and 3). Based on the history and clinical examination a provisional diagnosis of dentigerous cyst in

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relation to 21, 22 were given and AOT was considered under differential diagnosis.

Intraoral periapical radiograph, maxillary anterior occlusal radiograph, orthopantomogram, and blood investigations were advised. Intraoral periapical radiograph (Figure 4) showed a well-defined unilocular radiolucency with impacted 21. Maxillary anterior occlusal radiograph (Figure 5) revealed that the crown appeared to be completely embedded in the bone. Presence of well-defined radiolucency with sclerotic border surrounding crown of the unerupted tooth in relation to 21 at 1-mm below the level of cementoenamel junction extending anteroposteriorly from alveolar process of 21 region till the distal aspect of 26 and mediolaterally extending from apical third of 11 till alveolar process of 24. The internal aspect was moderately radiolucent and displacement of 11 was noted. Orthopantomogram (Figure 6) was also taken which showed similar findings. Based on these imaging findings, a radiographic diagnosis of dentigerous cyst in relation to 21 was given with a radiographic differential diagnosis as AOT was given.

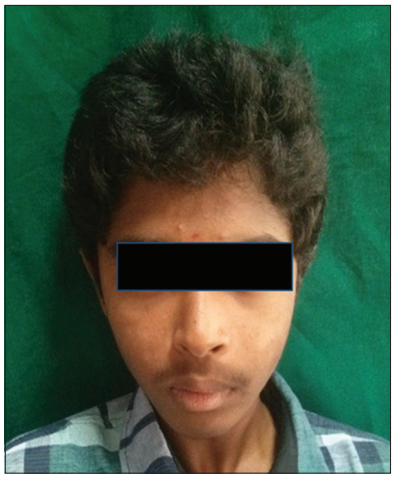

Figure 1: Frontal view

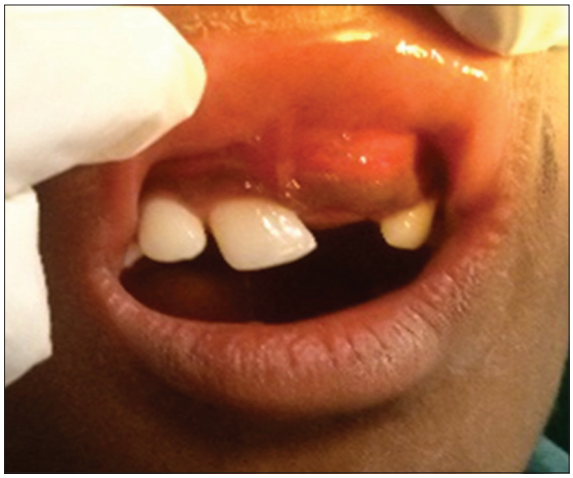

Figure 2: Intraoral labial view

In the treatment, surgical excision (Figure 7) was done and the specimen (Figure 8) was sent for the histopathological examination, which revealed cuboidal to columnar cells arranged in the form of nests and rosettes. Tubular

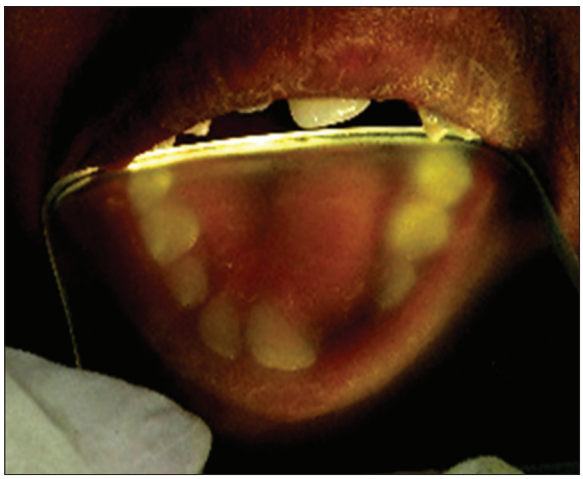

Figure 3: Intraoral palatal view

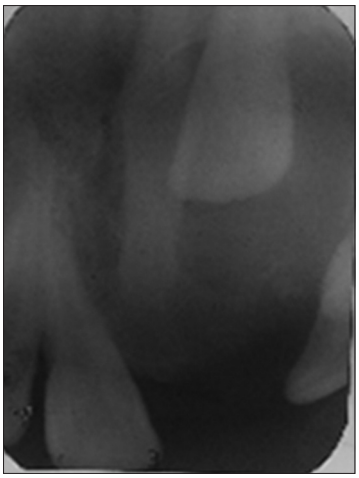

Figure 4: Intraoral periapical radiograph showing well-defined radiolucency with sclerotic border surrounding crown of the unerupted 21

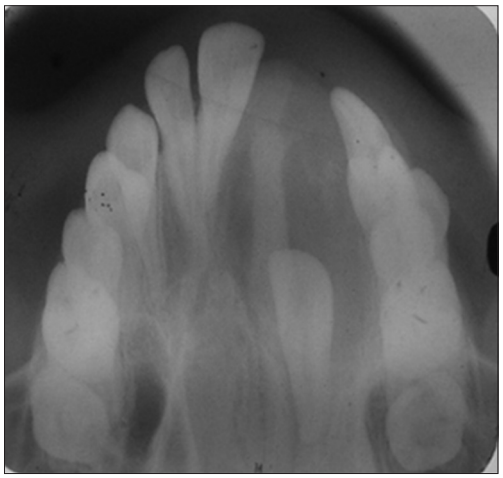

Figure 5: Maxillary cross-sectional occlusal radiograph showing the internal aspect which was moderately radiolucent with displacement of 11

appearance, solid areas, duct-like pattern, and whorled arrangement of cells is evident. Few cells were also arranged in a plexiform pattern and cribriform areas are also seen. At high magnification (Figure 9), sheets, nests of polyhedral cells along with ductal pattern lined by cuboidal to columnar cells at low magnification (Figure 10), sheets of epithelial cells along with ductal pattern. Hence, considering radiographic presentation and histopathological diagnosis, a final diagnosis of AOT in relation to 21 was given.

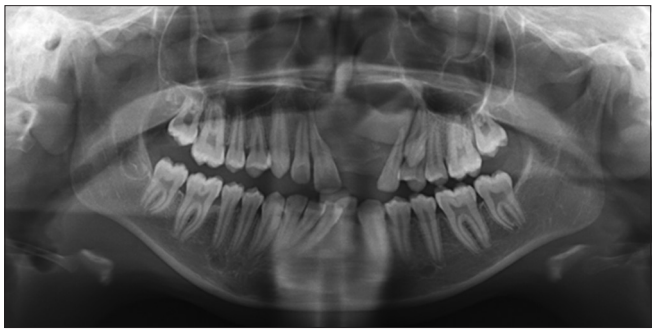

Figure 6: Orthopantomogram

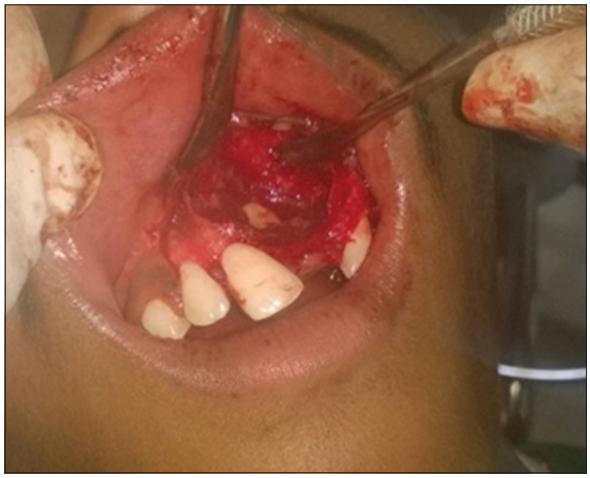

Figure 7: Intraoperative view

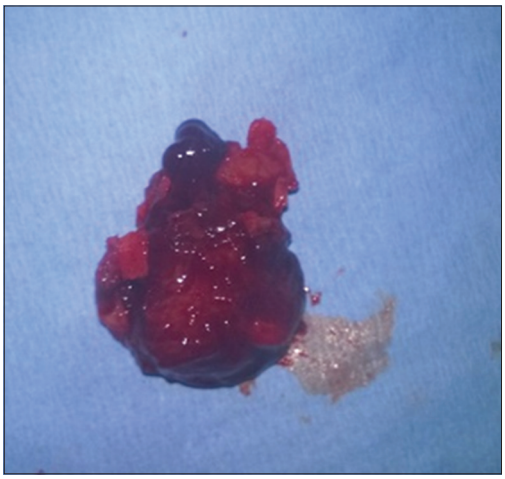

Figure 8: Specimen of surgically excised tissue

DISCUSSION

AOT is a benign, non-invasive odontogenic lesion showing slow growth. Over the years a variety of terminologies have been used to designate this odontogenic lesion like adenoameloblastoma, ameloblastic adenomatoid tumor, odontogenic adenomatoid tumor, pseudoadenoma adamantinum.[2] Philipsen and Birn proposed the name AOT (1969) and suggested that it should not be regarded as a variant of ameloblastoma because of its different behaviour.[3] AOT is also called "two-thirds tumor," because two-thirds occur in young females, two-thirds of these cases occur in the maxilla, two-thirds of these tumors are associated with unerupted teeth and two-thirds occur in canines.[6] It constitutes about 2-3% of all odontogenic tumors. AOT is divided into 3 variants by Philipsen *et al*., the follicular type (accounting for 73% of cases), which has a central lesion associated with an embedded tooth as seen in our case; the extra follicular type (24% of case), which has a central lesion and no connection with the tooth; and the peripheral

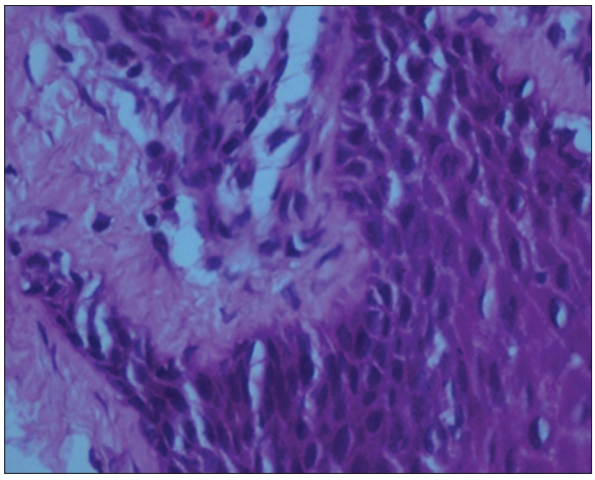

Figure 9: Histopathological picture in high magnification it showing sheets, nests of polyhedral cells along with ductal pattern lined by cuboidal to columnar cells

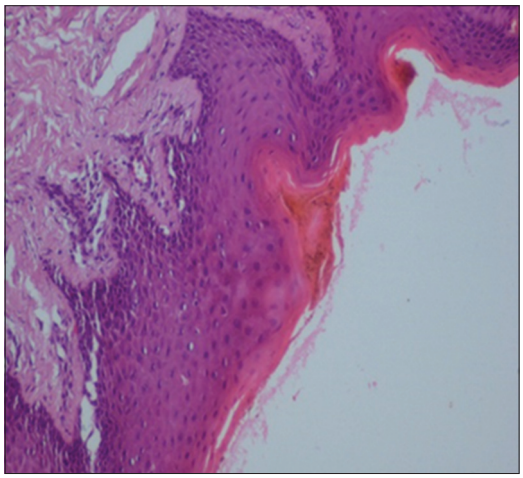

Figure 10: Histopathological picture in low magnification it showing sheets of epithelial cells along with ductal pattern

variety (3% of cases) which occurs primarily in the gingival tissue of tooth-bearing areas.[3]

The origin of AOT is believed to arise from an odontogenic source such as enamel organ, reduced enamel epithelium, dental lamina and their remnants. In long standing cases, the epithelial lining of the odontogenic cyst may transform into an odontogenic neoplasm - like an ameloblastoma or AOT.[7] 69% of AOT's are diagnosed in the second decade of life, as evident in our case. Incidence of this tumor in female to male ratio is at the rate of 2:1 and has a predilection for the anterior maxilla, in current case AOT occurred in male with anterior maxilla involvement.[4]

The lesions are usually asymptomatic and is often associated with cortical expansion. The involved teeth are commonly impacted and adjacent teeth may be slightly displaced. All the above-mentioned features were concurrent with our case. In general, the tumor does not exceed 1-3 cm in greatest diameter and usually occurs within the tooth-bearing areas of jaws and often associated with impacted teeth.[8]

The radiographic findings of AOT frequently resemble other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, ameloblastomas, odontogenic keratocysts and calcifying epithelial odontogenic tumor. In AOT, displacement of neighboring teeth due to tumor expansion is much more common. Root resorptions may also occur. In few cases, the peripheral lesions may also show erosions of the adjacent cortical bone. Intraoral periapical radiographs reveals radiopacities in AOT as discrete foci having a flocculent pattern within radiolucency even with minimal calcified deposits whereas panoramic radiographs often do not reveal. Approximately 78% of AOT shows calcified deposits. In our case, there were no calcifications seen.[5,9]

AOT is composed of spindle-shaped or polygonal cells forming sheets and whorled masses in a scarce connective tissue stroma. The characteristic duct-like structures are lined by a single row of columnar epithelial cells and the nuclei are polarized away from the central lumen. The lumen may contain amorphous eosinophilic material or it may be empty as seen in our case. Most of the AOTs show dystrophic calcification in varying amounts and forms within the lumina of duct-like structures and scattered among epithelial masses.[10,11]

Conservative surgical enucleation is the most suggested choice of treatment which was done in our case. The recurrence rate for AOT is rare and prognosis is good.[5] The present case has been on follow-up since 6 months after the surgery, no recurrence is noted.

CONCLUSION

We conclude that successive unerupted permanent teeth or persistence of deciduous teeth for a longer duration when associated with a swelling should always be suspected for odontogenic lesions particularly AOT should be considered under differential diagnosis.

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