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CASE REPORT

ADENOMATOID ODONTOGENIC TUMOR: A CASE REPORT

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is a distinct odontogenic tumor that is exclusively odontogenic epithelial in origin which accounts for about 3-7% of all odontogenic tumors, being benign (hamartomatous), noninvasive with slow but progressive growth (Batra *et al.,* 2005). It is predominantly found in young female patients, located more often in the maxilla in most cases (and) associated with an unerupted permanent tooth (Stafne, 1948). Treatment involves conservative surgical excision and the prognosis is excellent. Here we report a case of adenomatoid odontogenic tumor (AOT) in the left anterior maxilla in a young girl aged 16 years.

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INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is an uncommon benign epithelial lesion of odontogenic origin. Previously it was known as pseudo adenoameloblastoma by Dreibladt (1907) (Batra *et al.,* 2005). Later Staphne (1948) first recognized this as a distinct pathological entity. It constitutes about 2-3% of all odontogenic tumors (Stafne, 1948). The synonyms of AOT include adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantium and teratomatous odontoma. Philipsen and Birn (1969)suggested the name "adenomatoid odontogenic tumor", which is now widely used. Philipsen et al. divided AOT into three variants namely the follicular type, extra follicular type and the peripheral variety.The follicular type is a central intraosseous lesion associated with an impacted tooth, while extrafollicular intraosseous AOT has no relation with a unerupted tooth. The peripheral variant arises as a gingival fibroma or epulis attached to the labial, almost exclusively in the anterior maxillary gingiva. The central variant accounts for 97.2% and from which 73.0% of its type are follicular. The follicular variant (M: F ratio 1-1.9) is three times as frequent as extra-follicular type (Philipsen *et al.,* 1997). The follicular variant is diagnosed earlier in life (mean17yrs) than extra follicular type (mean 24yrs): 53.1%of all variants occur within the teen years (13-19yrs).

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A study has shown that follicular AOT was associated with one embedded tooth in 93.2%. Maxillary permanent canines account for 41.7% and all four canines for 60.1% of AOT-associated embedded teeth. The follicular variant of AOT is thought to be originating from the reduced enamel epithelium of the dental follicle. The origin of extra follicular remnants remains less clear (Jivan *et al.,* 2007). In the 2005 WHO classification, AOT was included under "odontogenic epithelium with mature, fibrous stroma without odontogenic ectomesenchyme" (Philipsen and Brin, 1969). Tumor appears as intra oral–extra oral swelling of maxilla and is sometimes called as "two thirds tumor" because it occurs in maxilla in 2/3 cases, around 2/3 cases in young females, 2/3 cases associated with unerupted teeth and 2/3 affacted teeth are canines (Marx and Stern, 2003). The lesion may contain only the crown of the tooth or whole tooth. It usually occurs in the anterior region of the maxilla (Neville *et al.,* 1995). Conservative surgical enucleation is the most suggested choice of treatment. Recurrence rate for AOT is exceptionally rare (Mutalik *et al.,* 2012). Here, we are presenting a case of Adenomatoid Odontogenic Tumor (AOT) in the left anterior maxilla in a young female.

CASE REPORT

A 16 years old girl reported to the Department of Oral Medicine and Radiology with a chief complaint of asymptomatic swelling in the upper left front tooth region since 5 months. Initially, swelling started in the left maxillary anterior region, which was small in size and gradually increased to present size. Swelling was asymptomatic and

history of trauma 5 years back while playing in school groundwas associated with it. There was no other significant history except the trauma. Medical and family history was not contributory. On extra-oral examination, mild facial asymmetry with the obliteration of the Naso-labial fold on left middle 3rd of the face was noted. Surface over the swelling was normal. Swelling was hard in consistency and non-tender on palpation. Ipsilateral left solitary submandibular lymph node was mobile, soft, and nontender on palpation (Figure 1). Intraoral examination revealed a solitary diffuse swelling in the anterior maxillary labial vestibular region extending from mesial aspect of 21 to distal aspect of 24, roughly oval in shape measuring about 1.5 cm × 2 cm in greatest dimension. The color of overlying mucosa was normal. On palpation, swelling was firm in consistency and non-tender (Figure 2). There was clinically missing 23 and over retained 63, 85. Based on the history and clinical examination a provisional diagnosis of Cystic lesion in the left anterior maxilla was given with the differential diagnoses as dentigerous cyst and Adenomatoid Odontogenic Tumor (AOT) was considered. Patient was subjected to routine radiographic examinations and hematological investigations. Intraoral periapical radiograph (Figure 3) showed a well-defined oval shaped unilocular radiolucency measuring about 2.5 x 3.5cm , surrounding crown of impacted 23 and extent 3 to 4mm below the level of cementoenamel junction (modify this line seeing the radiograph).

Figure 1: Extraoral view showing obliteration of nasolabial fold

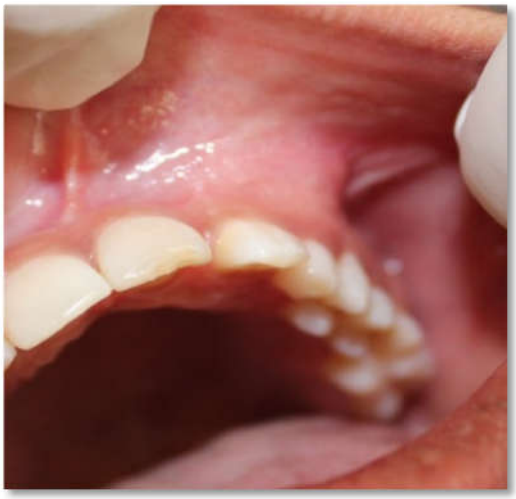

Figure 2. Intraoral view demonstrates the palatal aspect of the swelling

Maxillary anterior occlusal radiograph (Figure 4) revealed well-defined radiolucency which is oval in shape with sclerotic border surrounding crown of the unerupted tooth in relation to 23 at 3 to 4 mm below the level of cementoenamel junction extending anteroposteriorly from alveolar process of 23 region till the distal aspect of 24 and mediolaterally extending from apical third of 21 till alveolar process of 24 region. The internal aspect was completely radiolucent and displacement of 22 and 24 was noted. Panoramic Radiograph (Figure 5) was also taken which showed similar findings.

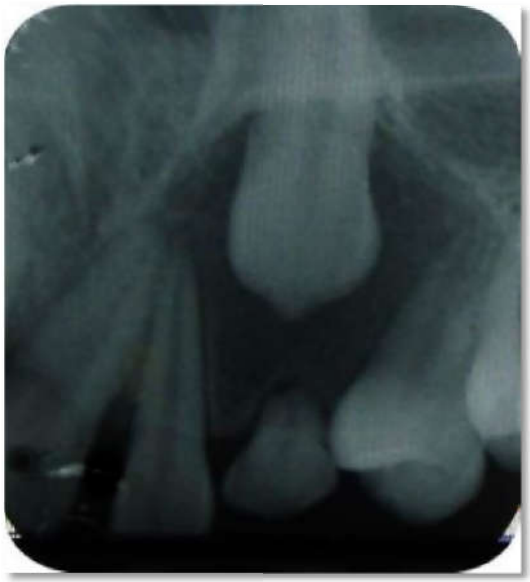

Figure 3. Intraoral periapical radiograph showing well-defined radiolucency with sclerotic border surrounding crown of the unerupted 23

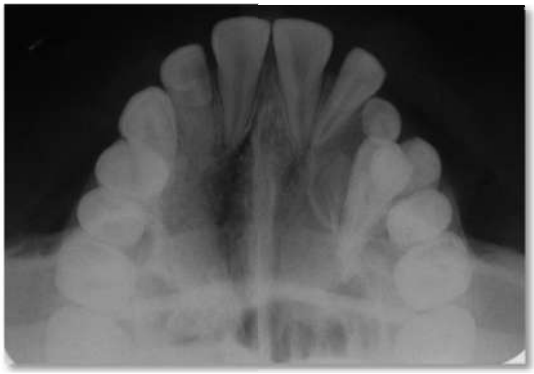

Figure 4. Maxillary cross-sectional occlusal radiograph showing the internal aspect which was moderately radiolucent with displacement of 22

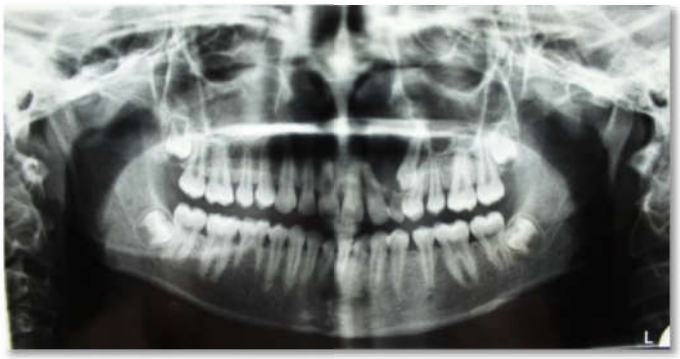

Figure 5. Orthopantomograph reveals Unilocular Radiolucency with an impacted 23

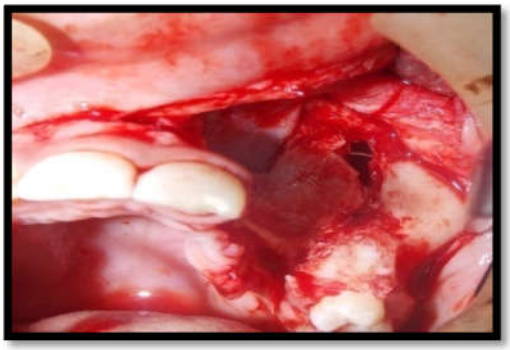

Figure 6. Intraoperative view

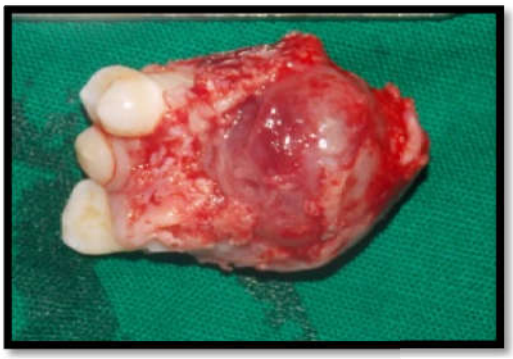

Figure 7. Photograph showing surgically removed specimen

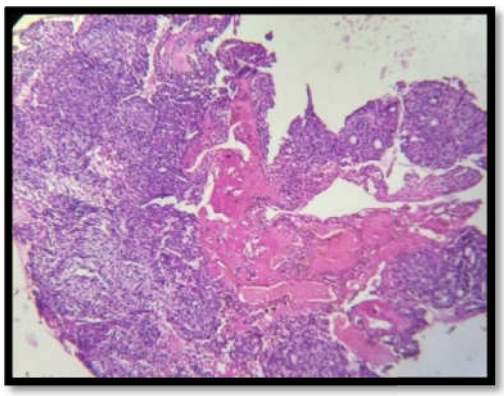

Figure 8 Histopathological picture in low magnification it showing sheets, nests of polyhedral cells along with ductal pattern lined by cuboidal to columnar cells

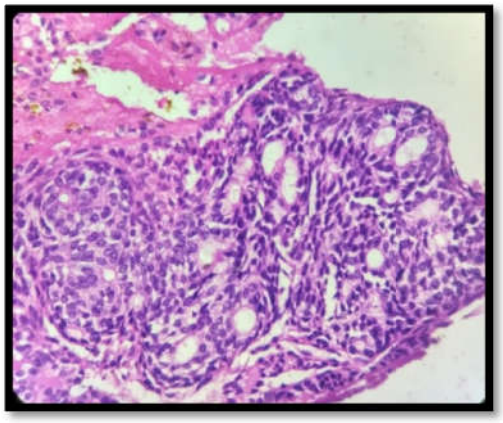

Figure 9. Histopathological picture in high magnification it showing ductal Lumina with varying size surrounded by columnar palisading epithelial cells darkly stained cells arranged in whorls and sheets of columnar cells throughout the fibrous connective tissue stroma

Based on these imaging findings, a radiographic diagnosis of dentigerous cyst was given with a differential diagnosis as AOT. Patient underwent Enbloc resection of the cystic lesion i.r.t 22 23 24 region with extraction of 22 63 23 24 and reconstruction using right iliac crest graft under general anesthesia (Figure 6), and the specimen (Figure 7) was sent for the histopathological examination, which revealed fibrous wall with solid nests and whorled nodules of epithelium cells. The cells were cuboidal spindle shaped and columar cells with polarized nuclei forming rosettes and duct like structures. Eosinophilic amorphous material seen the tumour cells and within duct like structures. Focal areas of calicification seen areas of bony tissue seen. At high magnification (Figure 8), sheets, nests of polyhedral cells along with ductal pattern lined by cuboidal to columnar cells at low magnification (Figure 9), sheets of epithelial cells along with ductal pattern. Hence, considering radiographic presentation and histopathological diagnosis, a final diagnosis of AOT in relation to left maxillary anterior region. The treatment was uneventful. After 3 months, Panoramic Radiograph showed the signs of healing of the surgical site. Patient is still under follow up with no signs of recurrence (Figure 10).

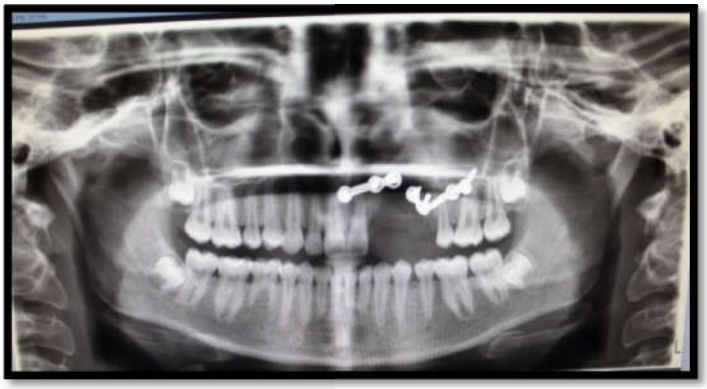

Figure 10 Orthopantomograph showed radiographic signs of healing of the surgical site

DISCUSSION

AOT is a benign, non-invasive odontogenic lesion showing slow growth. It is generally intraosseous, but can also occur rarely in peripheral locations (Philipsen *et al.,* 2002). Over the years a variety of terminologies have been used to designate this odontogenic lesion viz adenoameloblastoma, ameloblastic adenomatoid tumor, odontogenic adenomatoid tumor, pseudoadenoma adamantinum (Stafne, 1948). Philipsen and Birn proposed the name AOT (1969) and suggested that it should not be regarded as a variant of ameloblastoma due to its different behaviour. 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 variety (3% of cases) which occurs primarily in the gingival tissue of tooth-bearing area (Philipsen *et al.,* 1997). 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 (Swasdison *et al.,* 2008). This tumor is in the ratio of 2:1 with

female predilection and more common in anterior maxilla (Philipsen *et al.,* 1991), in current case AOT occurred in female with anterior maxilla involvement. The lesions are usually asymptomatic and are often associated with cortical expansion. The involved teeth are commonly impacted and adjacent teeth may be 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 area of jaw and often associated with impacted teeth (Marx and Stern, 2003). 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. 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 (Sandhu *et al.,* 2010). Enbloc resection of the cystic lesion i.r.t 22 23 24 region with extraction of 22 63 23 24 and reconstruction using right iliac crest graft under general anesthesia was done in our case. The recurrence rate for AOT is rare and prognosis is good. The present case has been on follow-up since 9 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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