



CASE STUDY

ADENOMATOID ODONTOGENIC TUMOR- A NONPAREIL CASE

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ABSTRACT

Adenomatoid odontogenic Tumor (AOT) tumor found to be a rare variety comprising of 3-7% of all odontogenic tumors. The most common location for AOT is anterior maxilla and very often confused with dentigerous cyst having similar radiological presentation. The tumor occurs in two forms central or intraosseous which is more common and peripheral being rare. Treatment of the tumor involves enucleation and curettage. We report a case of young female having AOT in left anterior maxilla with clinical, radiological, histopathological presentation and the management.

Key words:

AOT, Odontogenic tumors.

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INTRODUCTION

AOT is benign neoplasm having female predilection with most common occurrence in maxillary anterior region. The tumor presents in two forms central or intraosseous and peripheral. The tumor is also known as 'Two Third's Tumor' because about 2/3rd cases reported in maxilla, about 2/3rd cases are diagnosed in young females during the second decade, 2/3rd cases are found to be associated with impacted canine.

Case presentation

A 14 year old girl reported to department of oral medicine and radiology having complaint of Swelling in left maxillary region since 6 months. Swelling was slow growing and was not associated with pain. Extraorally single diffuse swelling seen on left side maxillary region extending from left ala of nose to 1.5 cm ahead of left ear lobule. Vertically it was extending from infraorbital margin till corner of mouth. No color change was noted of overlying skin on extraoral inspection. Intraorally (Fig. 1) swelling was present in maxillary left labial vestibule extending from 21 to 26 region causing buccal expansion and obliteration of vestibule. On palpation swelling was firm, nontender and smooth. Maxillary left deciduous canine seen

over retained with missing permanent canine. Orthopantomogram and PA waters radiograph (Fig. 2, 3) revealed a single well defined radiolucency surrounding impacted canine with radio opaque borders. Provisional diagnosis of dentigerous cyst was made, and complete enucleation of cyst (Fig. 4,5) was done along with impacted canine. Peripheral osteotomy was done, hemostasis achieved and primary closure was done using resorbable 3-0 vicryl. The H & E stained section (Fig. 6) showed epithelium and connective tissue. The epithelium showed tumor mass surrounded by connective tissue capsule at places. The epithelial cells are arranged in duct like structures. At places rosettes formation and ductal arrangement is seen. Duct like structures are formed by cuboidal to low columnar cells at the periphery and oval to polygonal cells in the center. The ducts and rosettes are surrounded by round to spindle cells. Eosinophilic material is evident in few duct like structures. Loosely arranged collagen fibers are seen interspersed with fibroblasts. The overall picture is suggestive of "Adenomatoid Odontogenic Tumor"

DISCUSSION

Herbitz from reported first AOT in 1915 and named it "admantoma." Later Philipson and Birn proposed the name "AOT" in 1969. (Rick, 2004) The tumor presents in two forms central or intraosseous as in our case and peripheral being less common variety. (Dhupar et al., 2016) Many terminologies being used to emphasize its cystic nature since its earliest

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description in 1905, which include “epitheliomaadamantinum,” “cystic adamantoma,” “adenoameloblastoma,” “cystic complex composite odontoma,” “ameloblasticodontogenic tumor,” “odontogenicadenomatoid tumor.” Marx and Stern, (2003) proposed that adenomatoidodontogenic cyst is would be more appropriate terminology since it often presents as a cystic lesion with intraluminalproliferation. (Belgaumi *et al.*, 2015; Marx and Stern, 2003) Though the origin of AOT is debatable issue few authors suggested that it could be derived fromepithelial remnants of dental lamina. (Raheel *et al.*, 2015) Male to female ratio of AOT reported is 1:1.9 the same was reflected in our case. (Rezvani *et al.*, 2015) Most common impacted tooth reported in AOT cases is maxillary canine as seen in our case. (Stafne, 1948) AOT (follicular variety) is very often misdiagnosed as dentigerous cyst since both shows well-defined unilocularradiolucency that surrounds the crown of an impacted canine. Aspiration reveals straw colored fluid which helps in differentiating dentigerous cyst from the solid tumor clinically and grossly follicular type of AOT sometimes extends apically beyond the cement enamel junction. (SumitMajumdar *et al.*, 2015) In our case radoilucency surrounding impacted canine was beyond cement enamel junction favoring the diagnosis of AOT. There have been many report wherein AOT found to be associated with dentigerous cyst. The odontogeniccyst epithelial lining may transform into benign neoplasms like AOT or ameloblastoma. (Jivan *et al.*, 2007) Histologically AOT can be seen as variety of cellular patterns.



Fig. 3. PA waters showing single well defined radiolucency in relation with impacted 23. No calcifications seen

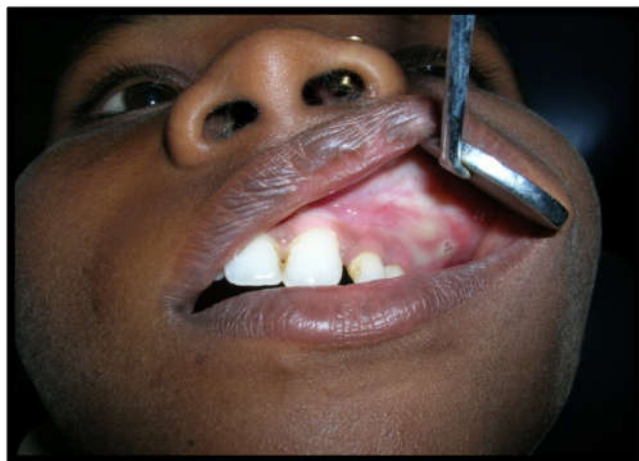


Figure 1. Clinical photograph showing intraoral swelling in left maxillary region



Fig. 4. Intraoperative photograph performing enucleation of tumor



Fig. 2. Orthopantomogram showing single well defined radiolucency with corticated border surrounding impacted 23

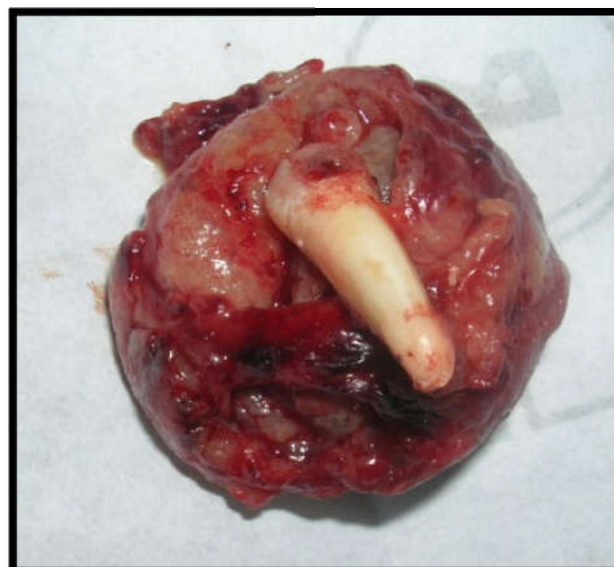


Fig. 5. Surgically enucleated tumor

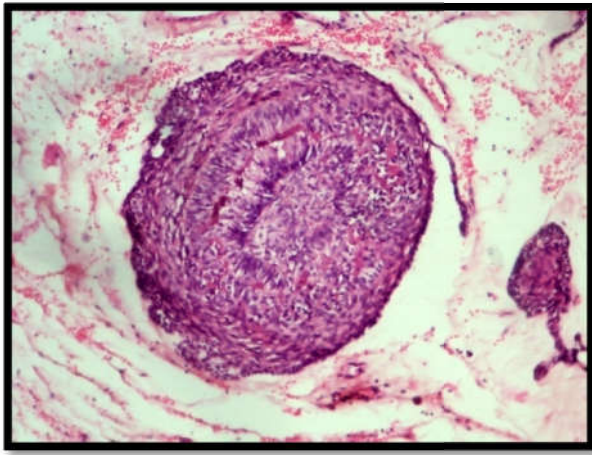


Fig. 6. Characteristic rounded and rosette form, lined by layer of polarized columnar or cuboidal epithelial cells giving adenomatoid appearance

Duct like structure lined by single row of columnar epithelial cells, polarized nuclei that are away from central lumen confirms diagnosis of AOT histopathologically. (Jindwani *et al.*, 2015) some cases reported extensive dystrophic calcification which was not seen in our case. Management of this tumor involves conservative enucleation and curettage. AOT is having low recurrence so enucleation and peripheral ostectomy provides success of this lesion. (Chhavi Jindal *et al.*, 2014)

Conclusion

The article presents a rare case of AOT. The case is important due to its cystic nature and varied histopathological presentation. Detailed studies at molecular level suggested on account of controversies related to the origin of the tumor.

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