

Adenomatoid Odontogenic Tumor – A case report

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Abstract

Adenomatoid Odontogenic Tumor is an Odontogenic tumor arising from enamel organ of dental lamina, predominantly found in young female patient in anterior maxilla. We report a case AOT in young male patient in relation with the premolar. CT scan and histopathological reports confirmed the diagnosis, patient underwent the Surgical Enucleation of the tumor and functional replacement was given after satisfactory healing of the site and further treatment has been planned for needful of the patient.

Keywords: Odontogenic tumor, Benign, Extra follicular.

Introduction

A tumor of Odontogenic epithelium with duct like structures and with varying degree 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 a masses in the wall of a large cyst. It is generally believed that the lesion is not the neoplasm.⁶ AOT was first

described by Steensland in 19052 and the first case was described by Dreyblatt in 19071. A variety of terms has been used to described this tumor like Adenoameloblastoma, Ameloblastic adenomatoid tumor, Adamantinoma, Epithelioma adamantinum or Teratomatous odontoma,³ until Philipsen and Birn in 1969 introduced the presently accepted nomenclature of adenomatoid Odontogenic tumor⁵ which was later also adopted by WHO (2005) in their “Histological typing of Odontogenic tumors, jaw cyst and allied lesion”.⁶ Origin of AOT is still unclear but most of the literature support its odontogenic source as it occurs exclusively within the tooth bearing areas and often found associated with embedded tooth.⁷AOT has cytologic features similar to that of enamel organ, dental lamina and reduced enamel epithelium or their remnants. It is also considered as a developmental outgrowth or a hamartoma, while some consider it as a major neoplastic growth of odontogenic epithelium.^{7,12} Adenomatoid Odontogenic tumor is a

relatively uncommon, benign and slow growing Odontogenic tumor. The tumor accounts for 3-7% of all Odontogenic tumors with the mean age of approximately 18 years and age range of 5-53 years showing marked predilection for occurrence of young females patients (Female to male ratio of 2:1).¹⁰ The tumor mostly occurs in maxilla (65%) than mandible (35%) and more frequently occurs in anterior part of maxilla. However, AOT cases have also been described in posterior maxilla and mandible, but rarely beyond the premolars. About 53% of AOT cases occur in anterior maxilla, while 9% occurs in maxillary premolar region and about 2% occurs in molar region.⁷ The present article report a rare case of AOT related to premolar unlike it has more frequency of occurrence in maxillary anterior region.

Case Report

A 14 year male reported to the Department of Pedodontics and Preventive Dentistry with the chief complaint of swelling in the upper left front region of jaw since 1 month. The swelling was not associated with pain, discharge, trauma or any other concomitant symptoms, on examination; extraorally the face was bilaterally asymmetrical due to dome shaped bony hard swelling on maxillary left side of the face. And on palpation extraorally the swelling was non tender, extending superio-inferiorly 1.5 cm below the lower eyelid to 0.5 cm above corner of mouth, antero-posteriorly from the ala of nose to 2 cm posteriorly, approximating 3cm×2.5cm in size. Intraorally, a diffuse pale pink, tense, shiny, single swelling was observed in left buccal vestibule with respect to 22 23 64 25 region. On palpation a bony hard swelling was extending superio-inferiorly from mucogingival junction to beyond the mucosa and antero-posteriorly from the distal of 22 to mesial of 26; which was non-fluctuant, non compressible and non reducible in nature.

Physiologic mobility was present with 64 and Egg shell crackling was felt in the 23 64 region.

The teeth present were,

7 6 5 4 3 2 1	1 2 3 D 5 6 7
7 6 5 4 3 2 1	1 2 3 4 5 6 7

The periapical radiograph showed the root resorption with 64, 23 was distally drifted. Snow driven appearance was appreciated with the distal aspect of 23 to mesial aspect of 25. Maxillary occlusal radiograph (cross-sectional view) showed the expansion of buccal and palatal cortical plates with distal aspect of 23 extending to distal aspect of 27 region. The Orthopantomogram showed the impacted 44 with well defined mixed radio-opaque radio-lucent lesion. The sinus floor was displaced upward due to expansion of cortical plates. The complete hemogram of the patient was within the normal limits. The Fine Needle Aspiration Cytology was negative. The Cone Beam Computed Tomography showed a single large well defined lesion present with impacted 44 approximately of size 27×28 mm in dimensions. Expansions of buccal cortical plates with thinning and perforations in some regions, whereas palatal cortical plates were thinned and non-perforated. The floor of maxillary sinus was pushed superiorly. The tumor mass was enucleated under general anesthesia, and the tissue was sent for histopathological evaluation, which showed, under scanner view epithelial and connective tissue stroma and thick fibrous capsule surrounding the nodules of epithelium. Under 10X magnification the Odontogenic epithelial cells were arranged in various patterns like ductal or tubular, rossete in sheets and net pattern. Mostly cells are spindle shaped and cells forming ductal pattern are cuboidal having polarized nuclei away from lumen. Under 40X magnification the rossete pattern of Odontogenic epithelial cells were seen. On follow-up

visits the fixed functional space maintainer with 44 and 45 (band and loop) was delivered after one year and patient is still on follow up visits after every 3 months.

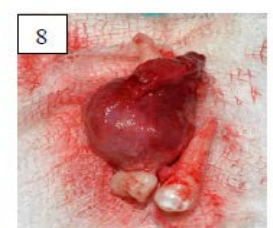
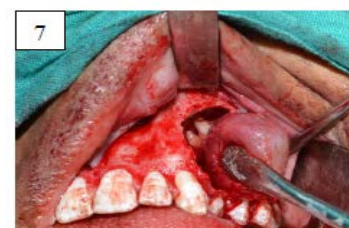
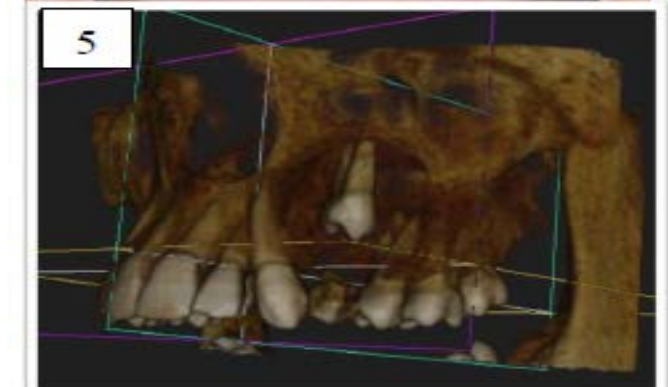




Figure 1. Pre operative picture showing swelling in maxillary left side of corner of mouth.

Figures 2,3,4. Pre operative intraoral photographs

Figure 5. Flap reflection and exposure of tumor mass.

Figure 6. CBCT 3D Reconstruction

Figure 7. Removal of tumor mass in toto.

Figure 8. Extraction of 64 and 25 along with the tumor mass

Figure 9. Sutures placed

Figure 10. Post operative pictures showing healing after 14 days.

Figure 11. Rehabilitation with fixed functional space maintainer.

Discussion

AOT has been previously described and classified as a variant of the ameloblastoma and named as Adenoameloblastoma, Adenoameloblastic odontoma, Pseudoadenomatous ameloblastoma, Cystic complex composite odontoma, Unusual pleomorphic adenoma like tumor, Ameoloblastic adenomatoid tumor, Odontoameloblastic tumor, Odontoameloblastic odontoma, Tumor of enamel organ, Ameloblastic epithelial tumor and Tumor connected to development cysts.⁸ Depending on the clinical and radiological findings AOT may be subdivided into variants such as-

A) Central (or intraosseous) it is further divided into two as Follicular and Extra follicular.

1) Follicular type (Dentigerous) in which the tumor is associated with the crown of an embedded tooth.

2) Extrafollicular type, in which tumor has no association with crown of an embedded tooth, which can be provisionally diagnosed as globulomaxillary or lateral periodontal cyst depending on the actual intraosseous localization of the lesion.

B) Peripheral variant (Extraosseous), in which the provisional diagnosis can be given as fibrosis or epulis.^{6,9}

Radiologically, AOT can be differentiated from dentigerous cyst as it surrounds the coronal as well as radicular aspect of involved tooth.⁸

The patient we described in this report was a male with premolar being the cause for AOT, presents the resorption of roots with 64, radiographically the radiolucency surrounds the crown and root portion of 24 (maxillary left first premolar). Macroscopically the tooth was embedded within the tumor mass. After the surgery the case is still under follow-up, the bony cavity has been observed healed optimally in the absence of any adjunctive therapy. With fixed functional space maintainer (band and loop) the premolar has been replaced with acrylic tooth and later the bone grafting and implant supported rehabilitation of first premolar is planned for needful of the patient.

Conclusion

It should be emphasized that the extrafollicular variant of AOT associated with premolars is very rare, along with this rarity of the tumor the slowly growing pattern, symptomless behaviour and radiograph helped to diagnose the tumor and post surgical histopathological evaluation confirmed the diagnosis.

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