

Large cystic Adenomatoid Odontogenic Tumor associated with impacted maxillary canine - a case report

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Abstract

Adenomatoid odontogenic tumour (AOT) is a benign, non-neoplastic lesion with slow progressing growth. It occurs in two forms intraosseous and peripheral forms. This lesion is commonly found in the anterior maxilla and is mostly associated with the impacted canine tooth. Here we report a case of large cystic AOT associated with an impacted canine appearing to be a dentigerous cyst grossly and radiographically. Histopathologically varied spectrum of the lesion were noticed, which have been documented and will help us to better our knowledge of AOT.

Keywords: Adenomatoid odontogenic tumor, Dentigerous cyst, Impacted tooth

Introduction

Adenomatoid odontogenic tumor (AOT) was first recorded in literature as epithelioma adamantinum by Steensland in 1905. Later, it was documented by Harbitz in 1915 as cystic adamantoma. But it was Philipsen and Birn who in 1969, proposed the widely accepted and currently used term Adenomatoid odontogenic tumor, which was adopted in the first edition of World Health Organization (WHO) classification of odontogenic tumors in 1971. In 2005, WHO defined, Adenomatoid odontogenic tumor as a tumor composed of odontogenic epithelium presenting different histoarchitectural patterns, embedded in a mature connective tissue stroma, and characterized by slow but progressive growth.⁽¹⁾

AOT is described as an uncommon benign odontogenic tumor with a relative frequency of 2.2-7.1%. It appears as an intraoral/ extraoral swelling in maxilla. It is sometimes referred to as "Two-thirds tumor" because two third of the cases occur in maxilla, two third of the affected are young females, two third cases are associated with unerupted tooth, and two third of the unerupted teeth were canines.⁽²⁾

As histogenesis of AOT is still uncertain, there has been long debate as to whether it represents anomalous hamartomatous growth, or is a true neoplasm. Though, it is currently accepted as a true odontogenic neoplasm. Rightfully called the "master of disguise", AOT has been known for its varied clinical and histoarchitectural patterns. This unique report of a large cystic AOT showing varied histoarchitecture is presented which may augur our understanding of the biology of the tumor.

Case Report

A 17 year old male patient reported to the clinical outdoor wing with a complaint of malaligned teeth.

Extra oral examination was insignificant. Intra oral examination revealed permanent dentition upto second molars in both the arches, except missing permanent left canine. Radiographic examination revealed an impacted permanent left canine completely surrounded by a large, well defined cystic lesion. A provisional diagnosis of an impacted canine associated with dentigerous cyst was given and surgery was advised followed by orthodontic treatment to correct the malalignment. After obtaining the patient's consent, enucleation of the cyst was done along with extraction of the associated impacted canine which was done under local anesthesia.

The gross specimen was brownish yellow in color, firm in consistency and was attached to the maxillary left canine, along with the cystic lining, measuring about 3 cm x 4.2 cm in dimension, the tooth was attached to the cystic lining in the cervical region (Fig. 1). The specimen had a gritty feel on cutting. The soft tissue bits were sent for routine histopathological examination. The histopathological examination of the specimen revealed cystic lining epithelium which was 2-4 layers thick, resembling reduced enamel epithelium (Fig. 2). The supporting connective tissue capsule comprised of bundles of collagen fibres lying parallel to the cystic epithelium. Certain areas revealed connective tissue capsule comprising of cuboidal to columnar cells arranged in the form of nests and rosettes (Fig. 3). In the same bit solid areas with duct like pattern, and tubular appearance was seen (Fig. 4). Few cells were also arranged in plexiform pattern. Foci of extravasated red blood cells was seen in few areas. Some amount of calcification, eosinophilic material was observed (Fig. 5). Correlating the histopathological features with the clinical and radiographic findings, a final diagnosis of cystic adenomatoid odontogenic tumor (AOT) was

given. The healing was uneventful and the orthodontic treatment is in progress.



Fig. 1: Photograph showing gross specimen with cystic lining attached to the tooth

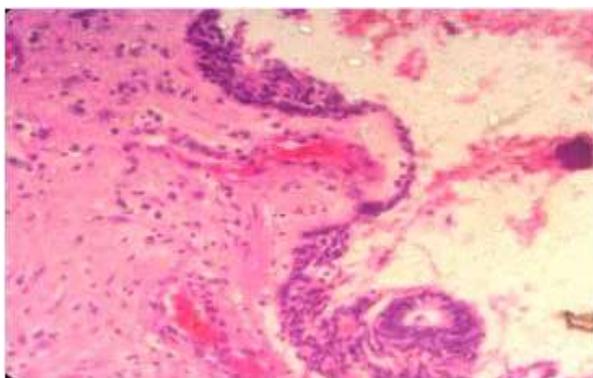


Fig. 2: Cystic lining comprising of 2-6 cell thick epithelial cells similar to dentigerous cyst (10x, H and E Stain)

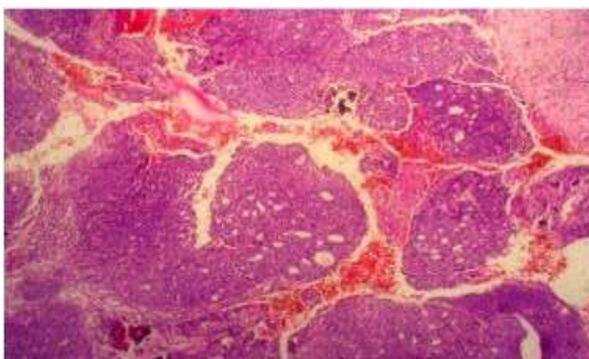


Fig. 3: Connective tissue capsule comprising of cuboidal to columnar cells arranged in the form of nests and rosettes (4x, H and E Stain)

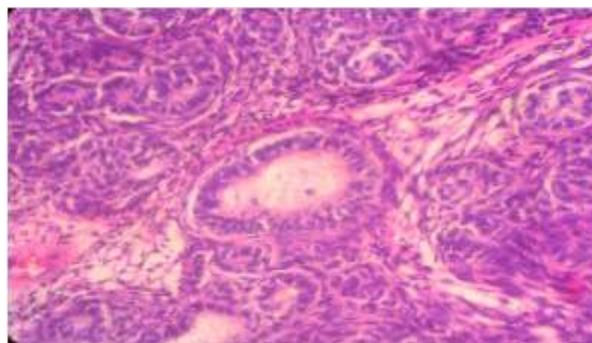


Fig. 4: Photomicrograph showing solid areas, duct like pattern, and tubular appearance (40x, H and E stain)

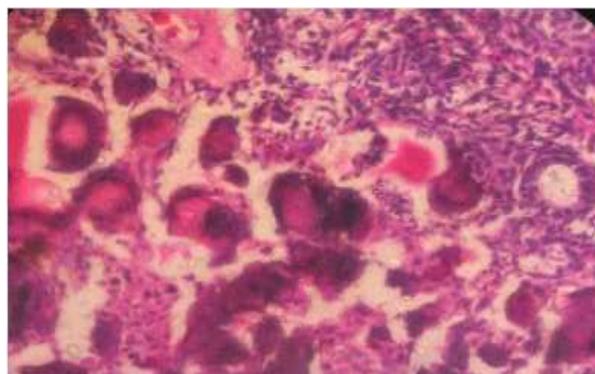


Fig. 5: Photomicrograph showing calcification and eosinophilic material (40x, H and E stain)

Discussion

AOT is a benign, non-invasive odontogenic lesion showing slow growth. Unal et al gave a list of all terms given to AOT in literature: adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum and teratomatous odontoma to name a few to describe AOT. It is generally intraosseous, but can occur in peripheral locations. AOT is mostly seen in young patients, especially in second decade of life, with mean age of 13.2 years and is uncommon in patients older than 30 years of age. Females are affected more than males with female:male ratio of 1.9:1. This is more marked in Asian population, with highest female incidence recorded in Sri Lanka (3.2:1). Maxilla is the most common site (Maxilla:Mandible=2.6:1). Anterior part of maxilla is commonly involved. An unerupted maxillary canine is the tooth generally associated with AOT.^(2,3)

AOT may be radiographically divided into two types: follicular or pericoronal and extrafollicular or extracoronal. Former is characterized as a well-defined unilocular radiolucent lesion surrounding the crown and is often part of the root of an unerupted tooth. The latter is a well-defined radiolucent lesion but located between, above or superimposed upon the root of an unerupted tooth. Minute variable shaped radiopacities are frequently found within the lesion.⁽³⁾ The follicular and extrafollicular variants are both intrabony and

account for approximately 96% of all AOTs of which 71% are follicular type. In fact 77% of all follicular types are diagnosed as dentigerous cyst.

Comparing the diagnostic accuracy between panoramic and periapical radiographs, Dare et al found that intra oral periapical radiographs allow perception of radioopacities having flocculent pattern within radiolucency even with minimal calcific deposits. Apart from that to differentiate between dentigerous cyst and AOT, in the former the radiolucency is never associated with the part of the root (always attached to the cervix) whereas the latter is always associated with the part of the root.⁽⁴⁾ In our case, the sclerotic margins were associated with the apex of the root but grossly the lining was attached at the cervix.

AOT is usually surrounded by well-developed connective tissue capsule. It may present as a solid mass, a single large cystic space, or as numerous small cystic spaces. The tumor is composed of spindle-shaped or polygonal cells forming sheets and whorled masses in a scant connective tissue stroma. Between the epithelial cells as well as in the center of the rosette like structures are amorphous eosinophilic materials and are called as “tumor droplets”. The characteristic duct like structures are lined by single row of columnar epithelial cells, the nuclei of which are polarized away from central lumen. Dystrophic calcification in varying amounts and in different forms is usually found in most AOTs within the lumina of the duct like structures, scattered among epithelial masses or in stroma.^(5,6) Our case showed all the classically described features of AOT along with the dentigerous cyst like lining epithelium.

In 1971, WHO defined this lesion as, “a tumor of odontogenic epithelium with duct like structures and with varying degrees of inductive change in the connective tissue. The tumour may be partly cystic and in some cases the solid lesion may be present only as masses in the wall of a large cyst. It is generally believed that the lesion is not a neoplasm”.⁽⁷⁾

Though the definition states the lesion as cystic very few cases have been reported having described cystic lining. Cystic presentation of AOT has been reported way back in 1915 by Harbitz as “cystic adamantoma”. The histopathological features seen in our case were both of dentigerous cyst and AOT. Grover et al also reported 10 cases of dentigerous cysts associated with AOT.⁽⁸⁾

Our case becomes unique because of the varied AOT features seen in the capsule of the cyst, grossly and histopathologically a challenging case.

Marx and Stern considered AOT as a cyst and not a tumor and coined the term Adenomatoid odontogenic cyst (AOC). According to them AOC does not arise from the follicle of the tooth crown but instead arises from HERS, which could explain the lining of the tooth being completely within the lumen rather than the tooth

root being inside the bone, which lead us to give the final diagnosis of cystic AOT.⁽⁹⁾

Philipsen et al have strongly argued in favor of the concept that AOT is derived from complex system of dental lamina or its remnants. The origin of AOT is controversial. However, most authors accept its odontogenic source. It occurs within the tooth-bearing areas of the jaws and is often found in close association of embedded teeth, having cytological features similar to those of components of enamel organ, dental lamina, reduced enamel epithelium or their remnants. Few support the idea that lesion is a developmental outgrowth or hamartoma while others consider it to be a neoplastic growth of odontogenic epithelium.^(8,9,10)

Conclusion

Adenomatoid odontogenic tumor (AOT) is a benign non-invasive odontogenic tumor, having mostly a slow and sustained growth pattern. It affects young individuals, has a female predilection and generally occurs in the second decade of life. AOT has been considered as a hamartoma rather than a true neoplasm because of its limited size, minimal growth potential and lack of recurrence. Even though enucleation and curettage for AOT is the most common treatment modality, accurate histological diagnosis is mandatory to avoid unnecessary mutilating surgery. Still the search for accurate classification and ideal nomenclature for AOT continues.

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