CASE REPORT

ADENOMATOID ODONTOGENIC TUMOUR - CYST OR TUMOUR??

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INTRODUCTION:
Adenomatoid odontogenic tumour is rare odontogenic lesion located either centrally within the jaws or peripherally in the soft tissue overlying the tooth-bearing area. Considerable debate exists about the histogenesis of this tumour; some investigators consider this tumour to be true benign, non-aggressive non-invasive neoplasms, whereas others categorized them as odontogenic hamartomas.1-4 In 2005 WHO defined this tumour as “Adenomatoid odontogenic tumour (AOT) is composed of odontogenic epithelium in a variety of histarchitectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth.”5

EPIDEMIOLOGY
AOT showed higher prevalence in Caucasian than the black Africans. This tumour is most often diagnosed in the second decade of life with a mean age 13.2 years and women are more commonly affected than men (2.3:1). Tumours involving maxilla is about twice as many affected in the mandible (2.6:1) and also preferably located in the anterior jaw the posterior area6, thus the tumour is sometimes referred to as two thirds' tumour.7

CLINICOPATHOLOGICAL CHARACTERS:
Clinically, it represents as a benign, painless, noninvasive, and slow-growing tumour that does not infiltrate the bone. It is often misdiagnosed as an odontogenic cyst, as its frequent association with an impacted tooth; more than 60 percent of the reported cases suggest the involvement of canine. Permanent incisors, premolars, molars, and deciduous teeth are rarely involved. A case of AOT associated with more than one tooth is also reported. Radiographically, AOT present as unilocular radiolucency with opacities and tooth displacement.1,2,3 This tumour shows three distinct clinico-topographic variants: 6

1. Intraosseous follicular type (Pericoronal):
Usually, located around the crown and often covers part of the root of an unerupted tooth (Envelopmental).

2. Extra follicular types (Extra coronal):
Commonly involves the following locations
   a. No relation to tooth structures
   b. Interradicular, adjacent roots diverge apically due to tumour expansion.
   c. Superimposed on root apex
   d. Superimposed on mid root level

3. Extraosseous peripheral epulis type (Extra
osseous): seen primarily in the gingival tissue of tooth-bearing areas and exhibit slight erosion of the bone crest. All the variants of AOT exhibit similar histopathological features. Usually these tumours are enclosed by a well-developed connective tissue capsule. Generally, it may present as a solid mass, a single large cystic space, or as numerous small cystic spaces and composed of spindle-shaped or polygonal cells arranged in sheets, whorled masses or in the form of ducts with in a scanty connective tissue stroma. Amorphous eosinophilic material is seen between the epithelial cells, as well as in the center of the rosette-like structure, known as hyaline ring. Duct-like arrangements of odontogenic epithelial cells are characterized single row of columnar epithelial cells exhibiting reverse polarity of the nuclei. Lumen may be empty or contain amorphous eosinophilic material. Varying amounts of dystrophic calcification in different forms are usually encountered within the stroma.1,2,6 Surgical management of this tumour includes enucleation along with the removal of associated impacted tooth with simple curettage. Conservative treatment is adequate as AOT behaves as locally invasive, well encapsulated, and is separated easily from the bone. The prognosis is excellent in majority of the cases, and previous literatures suggest the overall recurrence rate as low as 0.2%. In addition, larger osseous defects can be corrected using lyophilized bone and guided tissue regeneration.8 In this article we are reporting a rare case of Cystic adenomatoid odontogenic tumour occurring in the anterior maxilla and made an attempt to discuss the histogenesis and nomenclature of this rare lesion.

CASE REPORT:
A 12-year-old female presented with an asymptomatic, well-delimitated, 1.6 x 1.8 x 0.9 cm unilocular radiolucent tumour in the right anterior maxilla (Figure 1). On examination of the oral cavity, a mild buccal-palatal expansion of the anterior maxilla in relation to 11, 12, 13, and 14 was observed (Figure 2). Fine needle aspiration biopsy was performed, revealing straw coloured cystic fluid (Figure 3). Microscopic observation of cystic fluid showed odontogenic epithelial clusters along with few inflammatory cells, which were suggestive of odontogenic pathology. Correlating with clinical findings a provisional diagnosis of dentigerous cyst was made and the lesion was completely enucleated and was found to contain a cystic capsule adhering to a tooth (Figure 4). Microscopic observations revealed presence of cystic lining and supporting connective tissue capsule enclosing numerous islands of odontogenic epithelium (Figure 7).
Cystic cavity was lined by stratified squamous, non keratinized epithelium exhibiting proliferation into the underlying capsule in the form of nodules, cords and strands forming swirls of fusiform cells and ribbons of ameloblast-like cells. Duct-like arrangements of odontogenic epithelial cells as well as rosette like structures containing amorphous eosinophilic tumour droplets. These microscopic findings supported the final diagnosis of adenomatoid odontogenic tumour droplets. These microscopic findings supported the final diagnosis of adenomatoid odontogenic tumour (Figure 5, 6).

No recurrence was found upon 12 months follow-up after enucleation of the lesion.

DISCUSSION:

Origin of this tumour is debatable, however, due to its exclusive occurrence within the tooth bearing areas of the jaw and its cytological resemblance to the dental lamina and components of the enamel organ there is no disagreement that the AOT is of odontogenic in origin. According to Philipsen et al. all AOT have a common origin, probably arises from odontogenic epithelium of dental lamina complex or its remnants. The debate as to whether AOT is an anomalous hamartomatous growth or a true benign neoplasm is yet to solve. Immunohistochemical studies by few authors suggest the hamartomatous nature of this lesion indicating AOT is not a true neoplastic lesion. Odontogenic tumors and hamartomas constitute a wide variety of rare lesions that originate from odontogenic tissue and present with variable levels of differentiation. Understanding and identifying their precise nature is very difficult and results are inconclusive, thus makes it difficult to devise a nomenclature for this group of lesions. Reported literatures suggests the usage of various terminologies for AOT like lesions which includes, epithelial odontome by James and Forbes (1909), cystic adamantoma by Harbitz (1915), tooth germ cyst of the jaw by Wohlf of Omaha (1916), adamantinoma of the upper jaw by Ghosh (1934), epithelial tumours associated with developments cysts of maxilla by Stafne (1948), adeno ameloblastoma by Bernier and Tiecke, odontogenic adenomatoid tumor by Abrams et al (1968) and finally in 1969 Philipson and Birn first proposed the name adenomatoid odontogenic tumor. Later, this terminology was included in the initial edition of World Health Organization (WHO)’s classification of histological typing of odontogenic tumor, jaw cysts, and allied lesion (1971), from then till today numerous cases reports of this tumour have been published under the term adenomatoid odontogenic tumour.

First reported case of cystic presentation of AOT has been reported by Harbitz (1915); he preferred to use the term “cystic adamantoma”. Marx and Stern also suggested AOT as a cyst and not a tumour and further preferred the use of “adenomatoid odontogenic cyst” (AOC) as more appropriate term for AOT. According to them, the AOC does not arise from the follicle of the tooth crown but instead arises from HERS, which would explain the finding of the tooth being completely within the lumen rather than the tooth root being within a bony crypt. Coincidently, in the present case, presence of unilocular cystic lesion, fluid on aspiration, evidence of cystic cavity on and better prognostic features has to some extent support the terminology adenomatoid odontogenic cyst (AOC) as suggested by Marx and Stern.

Histogenesis of dentigerous cyst favours towards reduced enamel epithelium (REE) and not the dental lamina as Several studies have been performed in the past over cytokeratin (CK) profile of different variants of AOT and immunohistochemical analysis of CK in AOTs and found positive staining for CK5, CK17 and CK19. On contrary, classical AOTs expressed negative staining for CK5, CK17 and CK19 these findings are similar to follicular cyst and/or oral or gingival epithelium. On contrary, classical AOTs expressed negative staining for CK4, 10, 13 and 18. Crivelini et al detected the expression of CK 14 in AOT which suggests its origin probably from reduced enamel epithelium. In contrast, the present case showed tooth being embedded completely within the lumen and histological lining epithelium was not mimicking REE, which strengthens the Marx and Stern’s hypothesis of AOT originating from Hertwig’s epithelial root sheath (HERS). This concept was also supported by Sonal et al. they suggested all variants of AOT derived either from REE or HERS and begin as a cyst and later with the proliferation of nodules originating from the cystic lining fill up the entire lumen (solid type). Further, extra-
follicular and peripheral AOTs derive from the remnants of HERS (epithelial rests of Malassez).

**CONCLUSION:**
The exact histogenesis of AOT and all its variants still remains unclear. Though, AOTs represents in different clinico-topographic variants, still their clinical behaviour remains similar. In this report we made an attempt to hypothesize the new school of thought regarding the origin and nomenclature of AOTs. Further case reports and surveys of AOTs are necessary to define the precise relationship with the associated cyst.

**REFERENCES:**

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