**Mural Adenomatoid Odontogenic Tumour of Maxilla**

**ABSTRACT**

Adenomatoid odontogenic tumour (AOT) is a rare odontogenic tumour, it occurs more commonly in females with prevalence twice as compared to males. This paper reports a rare combination of AOT with dentigerous cyst in a 12 year old male patient involving the maxilla.

**Key words:** Adenomatoid Tumor; Adenomatoid odontogenic tumor; Male

**Introduction**

Adenomatoid Odontogenic Tumour (AOT) is a relatively uncommon, benign and slow growing tumour which is often misdiagnosed as an odontogenic cyst. AOT was first described by Steensland in 1905. Adenomatoid odontogenic tumour was described by Dreibladt, in 1907, as a pseudo-adenoma. In 1948 Stafne considered it a distinct entity, but it was classified by others as a variant of ameloblastoma. Philipsen and Birn proposed the name adenomatoid odontogenic tumour in 1969 and suggested that it not be regarded as a variant of ameloblastoma because of its different behaviour. This tumour was adopted by the World Health Organization (WHO) classification in 1971. In the latest edition of WHO classification of odontogenic tumours in 2005, AOT was classified into the first group of tumors (odontogenic epithelium without ectomesenchyme) instead of the second group (odontogenic epithelium with ectomesenchyme). Because of the absence of ectomesenchyme in immunohistochemical staining, dysplastic dentin, AOT is now considered the result of a metaplastic process rather than epithelial ectomesenchyme interaction. AOT is estimated to constitute about 2.2% to 7.1% of odontogenic tumors and the increasing number of reports on AOT points to the fact that the tumor develops more frequently than formerly expected. AOT is characterized histologically by the formation of ductlike structures with amyloid-like deposits and is a very uncommon cause of jaw swelling. AOT involves the anterior region of the maxillary bones, with a larger number of cases in females, in their second decade of life, and that may, sporadically, be associated with odontogenic cystic lesions. This paper reports a rare combination of AOT with dentigerous cyst in a male patient in the early second decade of life.

**Case Report**

A 12 year old male child was referred by a general dentist for evaluation of periapical pathology noticed in intra oral periapical radiograph. The medical history was insignificant and patient was in good general health. On clinical extra oral examination the swelling was observed on left side of anterior maxilla. Intraorally, the Swelling was present in upper left labial vestibule in 21-24 region, oval in shape, well defined margins of size 2x3 cm approximately with normal overlying mucosa (Figure 1). Expansion of labial as well as palatal cortical plate was appreciated. Over retained deciduous 62 and 63 with proximal caries with 63 and grade-II mobility with 62. The swelling was firm and non tender on palpation. Tooth missing were 22 and 23. Panoramic and occlusal radiographs revealed a well-circumscribed radiolucency of size 3x3 cm approximately around impacted 22 and 23 causing displacement of roots of 24 and 25 (Figure 2). On the basis of clinical-radiographic findings, the differential diagnosis was given adenomatoid odontogenic tumour, dentigerous cyst, odontogenic kerato-cyst, unicystic ameloblastoma, infected radicular cyst, calcifying odontogenic cyst and calcifying epithelial odontogenic tumour. Fine Needle Aspiration Cytology yielded clear straw colored fluid showed cholesterol crystals and desquamated cells suggestive of odontogenic cyst. The patient underwent an uneventful root canal procedure with 21, 24, 25, 26; followed by surgical removal under general anesthesia and healing was uneventful.

Macroscopically mass of 2.5x2.5 cm appears as soft, rubbery, spherical with a distinct fibrous capsule with root portion of teeth embedded inside the tumour mass. Upon gross sectioning, mass shows white to tan solid tissue with brownish granular material (Figure 3). Microscopically it revealed non-keratinized lining epithelium and showed proliferation into the lumen with spindle cells and columnar cells; with scattered duct like structures with lumina of varying size and lined by columnar cells formed whorls and rosette formation and filled in some areas with eosinophilic material (Figure 4).

**Discussion**

Adenomatoid odontogenic tumour is a slowly growing lesion, with a predilection for the anterior maxilla of young females. They are diagnosed in the second decade of life, and more than half occur during the teenage years. The female to male ratio for all age groups and variants is close to 2:1.5 In this case patient was male diagnosed in early second decade of life. The tumour has three clinicopathological variants, namely intraosseous follicular, intraosseous extra follicular, and peripheral. The follicular type is seen in 73% of all the AOT cases and is associated with an unerupted tooth as in our case, whereas the extra follicular type (24%) has no relationship with an impacted tooth, and the peripheral variant (3%) is attached to the gingival structures. In our case, the tumor was follicular intraosseous type and also found in the anterior region of the maxilla. Although larger lesions reported in the literature, the tumors are usually in the dimensions of 1.5 to 3 cm. Radiographically, they usually appear unilocular, may contain fine calcifications,
and irregular root resorption is rare. This appearance must be differentiated from various types of disease, such as calcifying odontogenic tumor or cysts. In CEOT borders may be ill-defined in addition small, thin, opaque trabeculae may cross the radiolucency in many directions. The differential diagnosis can also be made with unicystic ameloblastoma, ameloblastic fibroma, and ameloblastic fibro odontoma.

Appearance of internal bony septa is important for the identification of ameloblastoma in addition root resorption is commonly seen. The patient in the present report presented with no radiographic calcification and root resorption but showed hyperostotic borders with displacement of the adjacent teeth. It was also associated with an embedded tooth. AOTs macroscopically appear as a soft, roughly spherical mass with distinct fibrous capsule. Upon gross sectioning, the cut surface shows a variegated appearance with small areas of hemorrhage in grayish-white tissue. Small or large cystic spaces may be present and these may contain yellowish gelatinous material or blood stained fluid. In some cases, the tumour may be almost entirely cystic. A tooth or teeth may be embedded in the tumour or attached to the tumour mass.

According to the second edition of the WHO "Histological Typing of Odontogenic Tumors", AOT is defined as “A tumour 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.” The present case can be considered as AOT developed from the epithelial lining of dentigerous cyst as straw coloured fluid with cholesterol clefts are seen in cystic lesions only. No fine calcifications seen radiographically although lesion might started at least five years back therefore one can nominate it to be a mural AOT; as partly cystic nature of tumour. In this case clinical, radiographic, macroscopic and microscopic findings are consistent with descriptions of the lesions in the literature.

**Conclusion**

AOT occurring in male patient in early life is rare. Only careful diagnosis and the adequate interpretation of the clinical, radiographic and histological findings may be helpful in arriving at correct diagnosis. Conservative surgical approach is the treatment of choice as recurrence is rare.

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**References**


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