Adenomatoid Odontogenic Tumour with Calcifying Epithelial Odontogenic Tumour Component: A Rare Report of an Intricate Histomorphology
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Abstract
The adenomatoid odontogenic tumour is a benign, gradually developing, non-invasive tumour. It is recognised by its exceptionally notable duct-like structures. The present case of adenomatoid odontogenic tumour with calcifying epithelial odontogenic tumour component revealed areas of adenomatoid odontogenic tumour in the majority of the fields. Further detailed microscopic examination unveiled few calcifying epithelial odontogenic tumour like areas displaying excessive calcifications, giving rise to histological features of two odontogenic tumours in unison. This brings out with one of the unusual histopathological combinations reported in the literatures with a brief review of the literature.

Key Words: Adenomatoid Odontogenic Tumor, Calcifying Epithelial Odontogenic Tumour, Calcifications

Introduction
The adenomatoid odontogenic tumour (AOT) is a benign, gradually developing, non-invasive lesion. In 1969, Philipsen and Birn pointed out AOT as an unrelated entity from the solid or multicystic ameloblastoma after reviewing a total of 76 AOT cases. They introduced the term adenomatoid odontogenic tumour, adopted by the WHO in their “Histological Typing of Odontogenic Tumours, Jaw Cysts and Allied Lesions” as the present accepted nomenclature. It occurs in the intraosseous as well as in peripheral forms, the intrabony variants comprise of a follicular and an extra follicular types as appreciated in the radiographs.1

The WHO second edition “Histological Typing of Odontogenic Tumours” has defined the AOT as: A tumour of odontogenic epithelium with duct-like structures and with varying degrees of inductive changes 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. It occurs in uniformly unchanged manner also known as a “tumor of two-thirds” found two-thirds of female population in the second decades. Maxilla is more commonly involved with impacted canines, sometimes associated with dentigerous cysts.2 Fusion of odontogenic tumours is infrequently found in the literatures. They are specified by distinctive presentation of typical histological features of any recognised odontogenic tumours like calcifying epithelial odontogenic tumours, calcifying cystic odontogenic tumour, odontomas and other developmental odontogenic lesions.3,4

Calcifying epithelial Odontogenic tumour (CEOT) is recognised under many designations as ameloblastoma with calcification, calcifying ameloblastoma, malignant odontoma, adenoid adamantoblastoma, cystic complex odontoma and as a simple ameloblastoma variant ever since 1955.5 Because it is often linked with an impacted tooth, Pindborg suggested in an article from 1966 that the tumor arises from the reduced enamel organ.6

The present case report of AOT with CEOT component displayed AOT to a large extent. Detailed microscopic examination unveiled few CEOT like areas with excessive calcifications found scattered in the entire lesion. Thus presenting one more of the unusual combination reported in the literatures. This Paper gives attention to the histopathological observations of the two tumours.

Case Report
A 23 year old female patient visited a private clinic with complaints of mobility of upper right premolars with swelling in the anterior right maxillary region. It started as
asymptomatic swelling, later because of the teeth mobility patient visited the dental surgeon. Extra oral examination revealed incompatibility of the lips. On intraoral examination the soft tissue overlying the area was firm with evident swelling on the buccal and palatal sides. (Figure 1) Hard tissue observation revealed over retained right deciduous canine. Radiographs showed a large cystic lesion in relation to impacted right canine, resorption of premolar roots and flecks of calcifications. (Figure 2a) Computed tomography showed large expansile radiolucent lesion around impacted permanent canine with scattered radiopacities. (Figure 2b) A provisional diagnosis of AOT was considered.

Figure 1: Intraoral photograph of the patient shows palatal swelling in relation to upper right premolars.

Gross pathology of the enucleated biopsy specimen consisted of canine with a cystic cavity partly with a solid tumour mass attached beyond the cervical area of the tooth. The soft tissue was reddish brown in colour and firm in consistency measuring about 6x3x2cm. (Figure 2c) Histopathology displayed a fibrous connective tissue with odontogenic tumour islands made up of multinodular patterns of cuboidal and columnar odontogenic cells. Duct like structure, rosette patterns (Figure 2d & e) with eosinophilic materials, calcifications in the various forms were numerous. Clusters of polyhedral squamous epithelial cells displayed nuclear pleomorphism. (Figure 2f) Varied amount of dentinoid material with concentric layered deposition resembling liesegang rings, some of it had central basophilic areas were abundant. These small flecks of irregular calcification were seen together forming large continuous sheaths. (Figure 2g) Congo red staining revealed apple green birefringence under polarised light suggestive of amyloid like material. (Figure 2h) Few areas showed hyperchromatic oval cells as compressed islands in the fibrous connective tissue. (Figure 2i) All the distinctive features indicated the presence of the two odontogenic tumours AOT and CEOT. Final diagnosis, based on these findings, was AOT with a CEOT component.

Discussion
AOT is observed in 1% to 9% of all odontogenic tumours. Incidences are common among young females with unerupted maxillary canines. The present study followed the classical location of maxillary canine region reported in the literatures. Though the tumour displays an epithelial component, a notable pattern of typical duct-like structures, there is consistent occurrence of the tumour in a vast range of multiplicity. Foci of calcifications are usually scattered throughout the lesion. The number, size, and the degree of calcifications of these foci determine how the lesion presents radiographically.3

There is a clear delineation for the AOT to occur favourably in females, ratio of female to male range from 3.2:1 in Asian and 5.6:1 in African countries. The common variant of AOT is the follicular type (70.8%), as portrayed in the present case, and extra-follicular occurring in 26.9% of the cases, peripheral type is unusual seen in 2.3 % of all AOTs. Predictable site for AOT is found in the anterior maxillary region accounting for 88.0%. Combination of odontogenic tumours are a rare entity. According to Mosqueda-Taylor et al. CEOT-like areas in AOTs do not present as solid, infiltrative nests, as it appears in true CEOT. In addition, the CEOT-like areas in AOTs lack the typical pleomorphism that is found in the epithelial component of CEOT and their presence do not influence the biologic behaviour and growth potential of AOTs.4-7 In our case the histopathology showed typical areas of AOT, the classical ductal pattern which is unique to differentiate the AOT from any other odontogenic tumours. There were also quite extensive speckles of calcification spread all over the lesion with solid CEOT areas displaying quite evident nuclear pleomorphism among epithelial cells.

According to Philipsen and Nika, these calcifying materials are due to a metaplastic process, as odontogenic ectomesenchyme.
is not present in AOTs and therefore should not be interpreted as an induction phenomenon. In the earlier reported 24 cases of histological AOT/CEOT variants, calcifications were noted without dentin and cementum like hard tissues. According to Ledesma-Montes et al and Mosqueda-Taylor et al these areas are considered within the histomorphological spectrum of AOT. Ultrastructure analysis done by El-Labban and Lee in 3 cases of AOT confirmed the degenerative changes in 70.0–90.0% of the blood vessels found in the stroma because of the duplicated basal lamina. According to Philipsen et al these degenerative changes are also observed under light microscope.

**Figure 2:** The orthopantamograph shows radiolucency around the impacted upper right canine (a) with computed tomography reveals an expansile mass, with radiopacities around embedded tooth (b). Gross specimen demonstrates a spherical mass attached to the permanent canine beyond the cervical area (c). The hematoxylin and eosin stained photomicrographic sections under scanner view (d) show duct-like arrangements of tall columnar epithelial cells typical of AOT & clusters of polyhedral squamous epithelial cells of CEOT areas, whereas under low power view multinodular & rosette patterns of AOT (e) and CEOT areas showing epithelial cells with nuclear pleomorphism (f) along with irregular calcified structures (g). The photomicrograph of Congo red stained section demonstrating Amyloid like areas (h) and Van-Gieson stained section demonstrating hyperchromatic oval cells in compressed islands with fibrous connective tissue (x10).

The AOTs are frequently seen simulating other lesions like odontoma, CEOT, ameloblastic fibroma and other odontogenic tumours. The presence of cystic areas in AOTs has a close resemblance to odontogenic cysts, such as dentigerous cyst, has been reported in the literature. In the study done by Leon et al, 56.4% of AOTs exhibited cystic epithelium. Gadewar and Srikant found male prevalence over AOTs with cystic epithelium. The radiographic findings of AOT frequently resemble other odontogenic lesions, such as dentigerous cysts, calcifying odontogenic cysts, calcifying epithelial odontogenic tumours, keratocystic odontogenic tumour and ameloblastomas. In our case the diagnosis of a AOT was preferentially the first choice,
because of the involved impacted canine with the presence of radiopacities. The radiopacities in a lesion are always better compared with intraoral periapical radiographs, to a panoramic radiographs. 

An AOT often presents with fine radiopacities, which can be beneficial in the diagnosis. Sometimes a follicular AOT can resemble keratocystic odontogenic tumour and a unicystic ameloblastoma. But both these lesions are more common in the mandibular posterior regions unlike an AOT. Calciﬁying cystic odontogenic tumour needs to be distinguished from a follicular AOT as the location is quite similar and it may be found with an unerupted tooth. 

In conclusion, combined odontogenic tumours are uncommonly found and reported in the literatures. Our case of combined partly cystic and solid tumour of AOT/CEOT proves the complexity found among the odontogenic epithelium. Hence these tumours present with their own uniqueness in its histomorphological endeavours.

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