Hybrid Odontogenic Lesion of the Mandible: A Rare Case Report with Literature Review

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Abstract
A hybrid lesion is defined as being a "lesion showing the combined histopathological characteristics of two or more previously recognized odontogenic tumors and/or cysts of different categories." Adenomatoid Odontogenic Tumor (AOT) is relatively rare, and was previously described as a histological variant of ameloblastoma, but is now recognized as a separate entity by the World Health Organization, representing 3-7% of all odontogenic tumors. AOT is a slow-growing lesion that is thought to arise from odontogenic epithelium because of its predilection for tooth-bearing bone, with varying degrees of inductive changes in connective tissue. It is sometimes referred to as the "two-three tumor" because about two thirds occur in the maxilla, two thirds occur in young women, two thirds are associated with an unerupted tooth, and two thirds of these teeth are canines. Cases of maxillary involvement are more common, usually in the anterior region of the jaw. The present case describes the concomitant occurrence of AOT, dentigerous cyst and CEOT in the anterior portion of the mandible of a 13-year-old male patient. PAS staining was performed for confirmation of amyloid components, followed by Congo Red staining.

Introduction
Adenomatoid odontogenic tumor (AOT), an uncommon benign epithelial lesion of odontogenic origin, was first described by Dreibaldt in 1907 as Pseudoadenomaameloblastoma. Harbitz in 1915 reported it as "Adamantoma" and Bernier and Tiecke were the first to publish a case using the name "Adenoameloblastoma". Finally, the term "Adenomatoid Odontogenic Tumor" was proposed in 1969 by Philipsen and Birn(1).

According to the second edition of the World Health Organization’s "Histological Typing of Odontogenic Tumors", AOT is defined as "a tumor of odontogenic epithelium with duct-like structures and varying degrees of inductive changes in connective tissue." The tumor may be partly cystic, and in some cases, the solid lesion may be present only as masses in the wall of a large cyst(1).

Odontogenic tumors and cysts can occur at any stage of odontogenesis. The same embryonic origin supports a close interrelationship with several odontogenic lesions; therefore, it is not surprising that so-called hybrid tumors exist.

A combination of AOT and calcifying epithelial odontogenic tumor (CEOT) is considered to be representative of a hybrid odontogenic tumor, and is traditionally termed as combined epithelial odontogenic tumor(2).

As the histogenesis of AOT remains uncertain, there has been a debate as to whether it represents anomalous hamartomatous growth or is a true benign neoplasm. Currently, it is generally accepted to be a true neoplasm(3). While AOT is reported as a tumor in the histological sign out, that it represents a hamartomatous malformation adds a new dimension to its assorted histological architecture. This unique report with special emphasis on its histochotarchitectural spectrum may assist in our understanding of AOT biology(4).

Case Report
A 13-year-old male patient reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling in the lower left anterior region of the jaw for 6...
months. The swelling was painless, but was gradually increasing in size. The patient was apparently healthy, with no significant history related to the lesion and was not taking any medication for it.

Extra-oral examination revealed facial asymmetry with a single, oval, well-demarcated, non-tender, bony swelling in the mandibular left anterior region. Swelling extended from the corner of the mouth supero-inferiorly to the inferior border of the mandible, and 1 cm lateral to the chin antero-posteriorly to half the body of the mandible. The swelling was around 4×3 cm in size. There was no sign of numbness over the anterior region of the jaw (Fig. 1).

Intra-oral examination revealed firm swelling over the mandibular left anterior region extending from region 32 to 35, obliterating the labial and buccal vestibule with normal overlying mucosa. In addition, 74 was retained and was vital and immobile, while 34 was unerupted (Fig. 2).

Occlusal radiograph demonstrated that the lesion was expansile with labio-lingual expansion and there was thinning of the labial and lingual cortical plates.

Panoramic radiograph demonstrated a unilocular, round, radiolucent lesion of 3×3 cm, enveloping the impacted 34 in the left mandibular anterior region. The lesion was surrounded by a well-defined radio-opaque border with sclerosis in some areas. The lesion extended up to the lower border of the mandible in the canine and premolar region leading to thinning of the outer cortex. There was no root resorption, but displacement of 31, 32 and 33 was present (Fig. 3).

Provisional diagnoses made for the lesion was dentigerous cyst and the differential diagnosis made were unicystic ameloblastoma and AOT.

On aspiration with an 18-gauge needle, blood mixed fluid was obtained and was investigated as an inflammatory cyst. Blood examination revealed normal findings. The patient underwent surgery under local anesthesia, and 74 was extracted, while a full-thickness periosteal flap was raised from mandibular left 31 to 36 in the buccal vestibule. The buccal cortex was resorbed with a thin shell of bone in between. The lining of the cystic lesion was carefully separated from the mucoperiosteum and the lesion was enucleated along with the unerupted first premolar. The tumor cavity was irrigated with saline and betadine and was filled with gel foam, and the wound was sutured with 3-0 vicryl. Healing was uneventful and the patient has been
The excised mass was brownish in color and measured 3 × 2 × 2 cm. There was thick brownish fluid present within the mass. Histopathologic (H–E stain) examination revealed a cystic cavity lined by stratified squamous epithelium with multinodular proliferation of tumor cells that were columnar and cuboidal, and were arranged in ductular, convulated, tubular and rosette patterns. Sheets of polygonal cells with eosinophilic granular cytoplasm and giant nuclei with prominent intercellular bridges and calcification were observed (Figs. 4–8).

PAS and Congo red staining were positive. Histopathological diagnosis was thus confirmed as a dentigerous cyst converting to AOT and CEOT (Figs. 9, 10).
AOT is a slow-growing lesion, constituting only 3% of all odontogenic tumors with a predilection for the anterior maxilla (ratio 2:1) relative to the mandible, and is usually associated with impacted canine in young females in the second decade of life (5).

In our case the lesion occurred in the anterior mandible of a 13-year-old boy, which is unusual.

Rick et al., reviewed cases of AOT in association with dentigerous cysts and stated that although most central AOTs occur in a pericoronal relationship with an associated tooth. There is no way to be certain whether the lining of an associated cyst represents a true dentigerous cyst or a secondary cystic change within the AOT (6). In this case, AOT and dentigerous cyst were seen in the same lesion.

When we consider the etiopathogenesis of such a tumor, the origin of AOT is controversial. Some believe that they originate from the odontogenic epithelium of a dentigerous cyst. Santos et al. reported a case of AOT developing in the fibrous capsule of a dentigerous cyst, while Garcia-Pola et al., described the proliferation of an AOT in the epithelial border of a dentigerous cyst (7, 8).

All cases of AOT associated dentigerous cyst reported by John et al., and Garcia-Pola et al. have in common a unilocular radiolucency, while an AOT reported by Tajima et al. was seen as a radiopaque mass (8–10).

All of these occurred in the maxilla and only two were associated with the maxillary sinus. Of these, all except two occurred in the posterior region of the jaw and all were associated with impacted teeth, of which the most common tooth involved was the canine.

Common neoplastic cases, such as ameloblastoma, CEOT, ameloblastic fibroma and ameloblastic fibroodontoma are easily differentiated histologically. CEOT shows larger and...
more numerous calcifying spherules within the eosinophilic cytoplasm of large cells, along with smaller cells with hyperchromatic nuclei. Amyloid-like eosinophilic material is also present. Areas of CEOT-like tissues have been described in classic AOT (11).

Rashmi et al. reported a case showing a cellular pattern consisting of nodules of polyhedral, eosinophilic epithelial cells with squamous appearance and exhibiting well-defined cell boundaries and prominent intracellular bridges (5).

These islands may contain pools of amorphous amyloid-like material and globular masses of calcified material (thus suggesting a combination of CEOT and AOT) (5).

The most interesting finding in our case was that the majority of the above-mentioned histomorphologic patterns existed in one particular case. Calcification was seen in the form of irregular masses, leisegang rings, and spheroidal and globular forms. Although the tumor is odontogenic in origin, the reason for the occurrence of the ductal architecture remains uncertain. Few authors believe it to be due to a cystic change in the follicles of tumor islands or an attempt to form glandular tissue, as the origin is from basal cells of the oral epithelium, which possess multiple differentiation capacity. Moreover, the occurrence of all of these patterns could simply represent caricatures of the enamel organ itself. Of significant interest would be the occurrence of a CEOT-like area in AOT, which was believed to be an altered phenotype in certain parts of the tumor. This hypothesis was further supported by an immunohistochemical analysis using a panel of cytokeratin markers and vimentin, wherein the CEOT-like areas showed negative expression to CK 19.

It has even been suggested that these areas are the normal histoarchitectural spectrum of AOT (4).

The combination of two odontogenic tumors is a rarely reported finding. Combined or complex odontogenic tumors are lesions characterized by the synchronous presentation of typical histological features of two odontogenic tumors. The present case is a AOT-CEOT combined tumor, and has been reported previously (12).

More than 10 cases of combined AOT and CEOT (Pindborg tumor) have been reported since 1983. In 1983, Damm et al. presented the first 2 cases of combined epithelial odontogenic tumors, which contained areas diagnostic for both AOT and CEOT. The histological features, histogenesis and suggested treatment were discussed in their report at that time. The third case of the same previous combination was reported in 1986. This was followed by one additional case in 1987, 5 cases in 1991, 2 cases in 1993, one case in 1994, and one case in 1996. The most recent case of the previous combination was reported by Mosqueda-Taylor et al (13, 14).

Our case also presented as a unilocular radiolucency in the anterior region of the jaw and was associated with an impacted tooth, and the lesion was a histologically typical dentigerous cyst proliferating towards the luminal side to form typical AOT-and CEOT-like structures, which corresponds to the above-mentioned cases. However, our patient was male and most of the above lesions occurred in female patients, and the present lesion manifested in the mandible.

This case is of specific interest because of the vivid histological architecture present (dentigerous cyst converting to AOT and CEOT), and the involvement of an impacted mandibular first premolar.

References