Adenomatoid Odontogenic Tumor (follicular type) of the Mandible: A Case Report and Review of the Literature

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Introduction

Adenomatoid odontogenic tumor (AOT) is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland in 1905 (1). The name ‘adenomatoid odontogenic tumor’ was proposed by Philipsen and Birn (2) in 1969. However, a variety of terms has been used to describe this tumor. Unal et al. (3) produced a list containing all nomenclatures for AOT reported in the literature. Many different names, such as adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum, and teratomatous odontoma, have been used to describe the lesion now called AOT. This term was adopted by the World Health Organization (WHO) in 1971 (4). In 1999, Philipsen and Reichart (5) presented a review based on reports published before 1997 that showed some interesting aspects regarding the epidemiological data on this tumor. The current WHO classification (2005) of odontogenic tumors defines AOT as being composed of odontogenic epithelium in a variety of histarchitectural patterns, embedded in a mature connective tissue stroma, and characterized by slow but progressive growth (6).

There are three variants of AOT: follicular, extrafollicular, and peripheral. The follicular type is a central intrabony lesion associated with an unerupted tooth; this accounts for about 70% of all cases. The extrafollicular type is also an intra-osseous lesion, but unrelated to an unerupted tooth, and represents 25% of all AOTs. The peripheral type is a rare form that arises in the gingival tissue. All three variants have the same histological aspects and clinical behavior (7, 8).

Case report: A 20-year-old man reported to the Department of Oral Medicine and Radiology with an asymptomatic swelling in the anterior mandible that began 2 years earlier. The swelling was insidious in onset and gradually progressed to attain the present size. The patient gave a history of a missing tooth in the same region. There was no history of trauma. The lymph nodes in that vicinity were not palpable. Mouth opening was normal with no deviation on maximum opening. The patient’s medical, surgical, and dental histories were not significant.

Clinical examination: Extraoral examination (Fig. 1) revealed diffuse swelling in the mandibular symphysis and left parasympysis, measuring approximately 4 × 2 cm, roughly oval in shape, with ill-defined borders, a smooth surface, and normal skin over the swelling. On palpation, there was no rise in local temperature, no tenderness, the swelling
 Expansion of tissue and extended into the alveolar processes, disrupting the usual orientation of the anterior teeth. The left mandibular canine, which was present within the lesion, had been displaced considerably toward the contralateral side.

On the basis of the clinical and radiographic findings, the differential diagnoses of dentigerous cyst, adenomatoid odontogenic tumor, calcifying odontogenic cyst, calcifying epithelial odontogenic tumor, odontogenic keratocyst, and unicystic ameloblastoma were considered.

The patient underwent surgery with local anesthesia. A mucoperiosteal flap from the right to the left premolar was reflected to expose the labial aspect of the tumor. The labial cortex was very thin and had several areas of complete resorption. The tumor was enucleated along with the impacted lower permanent canine (Figs. 5 and 6). The areas between the roots of
the involved teeth were curetted well. The cavity was packed with Gelfoam® absorbable hemostatic gelatin sponge and the flap was sutured in place. Healing was uneventful and there was no evidence of recurrence 1 year after the surgery.

Histopathological examination revealed sheets of polygonal cells throughout the fibrous connective tissue stroma (Fig. 7a, 7b, 7b'). The ductal lumina were surrounded by columnar epithelial cells and filled in some areas with eosinophilic material. In other places, amorphous calcified material was present. The histopathological report confirmed the diagnosis of intraosseous follicular adenomatoid odontogenic tumor.

Discussion

AOT is a relatively slowly growing lesion seen approximately twice as frequently in the anterior maxilla than in the mandible. In our case, the AOT was in the anterior mandible. It is usually present in younger individuals in the second decade of life, with a female-to-male ratio of 2 : 1 (9). The prevalence of AOTs is 1.2% in Caucasians and 9% in Negroid patients (10). The lesions are typically asymptomatic,
but growth of the types with central lesions results in cortical expansion, as in the case reported here. The involved teeth are commonly impacted and adjacent teeth may be slightly displaced (11). Unerupted first and second molars are rarely involved, nor are deciduous teeth. Root resorption is a less common finding. If the lesion is large, it can cause a painless hard swelling, as in the case reported here.

A diagnosis of AOT should be considered in the differential diagnosis of corticate radiolucencies with small radiopaque foci, especially in teenagers and young adults. For cases in which the lesion appears to surround an unerupted tooth and has no radiopaque component, a dentigerous cyst may also be considered in the differential diagnosis. However, an AOT often appears to envelop the crown as well as the root, whereas dentigerous cysts do not envelop the roots (12–15). Radiographically, they usually appear unilocular, may contain fine calcifications, and irregular root resorption is rare. This appearance must be differentiated from various types of diseases, such as calcifying odontogenic tumors or cysts. The differential diagnosis can also be made with ameloblastoma, ameloblastic fibroma, and ameloblastic fibro-odontoma (16, 17).

The patient in the present report presented with no root resorption, but displacement of the adjacent teeth. The lesion was also associated with an embedded tooth. Radiographically, it was easily differentiated from dentigerous cyst, which usually occurs as a pericoronal radiolucency.

The origin of AOT is controversial (18). Because of its predilection for tooth-bearing bone, it is thought to arise from odontogenic epithelium. WHO has described the histologic features of the tumor as follows: “A tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive changes in the 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. It is generally believed that the lesion is not a neoplasm.” The histologic appearance of all variants is identical and exhibits remarkable consistency (19). At low magnification, the most striking pattern is that of various sizes of solid nodules of columnar or cuboidal epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue. The tumor may contain pools of amyloid-like material and globular masses of calcified material. Our case was consistent with the common features reported in the literature (16, 17). Immunohistochemistry is recommended for research purposes but not as a routine tool to establish the diagnosis of odontogenic tumors, including AOT (20).

An earlier report pointed out that the AOT phenotype is characterized by a cytokeratin profile resembling follicular cysts and gingival epithelium (9). This comparison was based on the knowledge of AOT immunoreactivity for cytokeratin subtypes,
which shows some zonal differences within neoplastic nodules. The tumor is well encapsulated and shows an identical benign behavior. Therefore, conservative surgical enucleation produces excellent outcome without recurrence.

Conclusion
A case of intraosseous follicular AOT is presented with clinical, radiological, and histological features, in which the rare and unique features were the swelling in the mandibular anterior region and bi-cortical expansion of the jaw. It is necessary to include AOT as one of the differential diagnoses for tumors involving the anterior mandible that are associated with unerupted or missing teeth.

References