

Mandibular adenomatoid odontogenic tumor: A report of an unusual case

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Abstract

Adenomatoid odontogenic tumor (AOT) is a slow-growing, benign tumor that appears in the anterior portion of the jaws and more frequently, the anterior maxilla usually in association with the crowns of unerupted teeth. Most of the tumors are diagnosed in second decade of life. A rare case report of AOT associated with an impacted right mandibular first premolar in a 24-year-old female is reported.

Keywords: Adenomatoid odontogenic tumor, follicular, impacted, mandible

Introduction

Adenomatoid odontogenic tumor (AOT) is a benign odontogenic lesion, which, it is hypothesized, develops from the enamel organ, dental lamina, reduced enamel epithelium, or their remnants.^[1-3] It was first described by Dreibaldtin in 1907 as pseudo-adenameloblastoma.^[4] Unal *et al.* produced a list of nomenclatures for AOT reported in literatures.^[5] Terms such as adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum, or teratomatous odontoma have been used before to define the lesion which is currently known as AOT. It was Stafne in 1948 who considered it as a distinct entity.^[6] The term AOT was proposed by Philipsen and Birn in 1969. Shortly thereafter, the latter term was adopted in the initial edition of the World Health Organization's (WHO) Histological Typing of Odontogenic Tumors Jaw cysts and Allied Lesions in 1971.^[7] AOT accounts for 2.2 to 7.1% of all odontogenic tumors, which gives it a ranking of fourth or fifth among the odontogenic tumors. Although it is generally believed that AOT is a hamartoma rather than a neoplasm, the lesion sometimes exhibits aggressive behavior. There are

three variants of AOT, the follicular type (accounting for 73% of cases), which has a central lesion associated with an embedded tooth; the extrafollicular type (24% of case), which has a central lesion and no connection with the tooth; and the peripheral variety (3% of cases).^[8] Both types of central intraosseous tumors produce a corticated radiolucency, sometimes with radiopaque specks. The follicular type is usually initially diagnosed as a dentigerous or follicular cyst. The extrafollicular type presents as a unilocular, well-defined radiolucency found between, above, or superimposed on the roots of erupted teeth and often resembling a residual, radicular, globulomaxillary, or lateral periodontal cyst. The peripheral type usually presents as a gingival swelling, located palatally or lingually relative to the involved tooth.^[2] This case report describes an unusually large follicular AOT in the mandible, illustrates the clinical, microscopic, and biological features of the tumor, and emphasizes the importance of the relation between the dental follicle and the tumor tissue. The article also provides a refresher for general dental practitioners about diagnostic aspects of this tumor.

Case Report

A 24-year-old female reported to the Department of Oral and Maxillofacial Surgery with chief complaint of pain and swelling over right mandibular region since 6 months. Intraoral examination revealed a firm swelling extending from right mandibular central incisor to first molar of the same side with obliteration of buccal vestibule. The overlying mucosa was ulcerated and pink in color. There was no paraesthesia over mandibular region. On aspiration, straw-colored fluid was obtained. Lateral oblique radiograph was taken, which revealed a well-defined unilocular, corticated radiolucency associated with impacted first premolar which extended almost to the apex of the tooth [Figure 1]. On the basis of clinical and radiographic findings, the differential diagnosis of dentigerous cyst, unicystic ameloblastoma, odontogenic keratocyst, and AOT was made.

The mass was enucleated completely along with embedded premolar under local anesthesia and submitted for histopathological examination.

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Gross examination of the tissue revealed a mass of 4 × 5 × 3 cm in size [Figure 2]. The mass was attached to the apical third of the root [Figure 3]. Corresponding to the gross, sections of the solid tissue contained histological characteristics of AOT. The tumor was composed of nodules of various sizes, consisting of cuboidal or columnar epithelial cells that formed nests, whorls, and rosette-like structures [Figure 4]. Foci of eosinophilic hyaline droplet material and calcification were also observed [Figure 5]. The connective tissue stroma was loosely arranged and contained thin-walled congested vessels [Figure 6]. Duct-like structures were located in many of epithelial nodules and these structures were lined by similar columnar cells [Figure 7]. Overall features were diagnostic for an AOT.

Discussion

AOT is a benign, slow-growing, epithelial tumor showing an intraoral location which manifests as a painless mass. Patients

are usually referred to dentists with complaints of impacted teeth within the lesion, which is usually a canine. The lesion may be associated with coronal portion of the tooth or the tooth in toto.^[1]

AOT usually occurs in the anterior region of the maxilla, but it may also arise in the mandible. In our case, the lesion was in the posterior mandibular region and contained an enclosed first premolar tooth. A review of English literature revealed twenty documented case reports. AOT is most commonly found in the second decade of life but in our case, it was found to be in third decade of life.^[2]

Radiographically, this lesion usually surrounds an unerupted tooth and is seen as corticated radiolucency with small radio-opacities, but there are cases where the lesion has no radio-opaque component, as in our case.^[3]

However, an AOT often appears to envelope the crown as



Figure 1: Lateral oblique radiograph showing well-defined unilocular radiolucency around impacted first mandibular premolar



Figure 2: Gross examination of the lesion measuring 5 × 4 × 3 cm in size along with thickenings on tumor mass



Figure 3: Cut section of the tumor shows relation of the tooth to the lesion

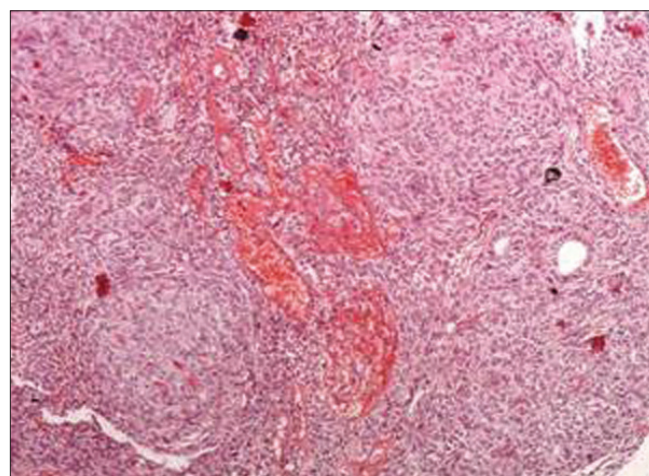


Figure 4: Tumor nodule composed of spindle-shaped or cuboidal epithelial cells forming rosette, duct, and whorls-like pattern (×10)

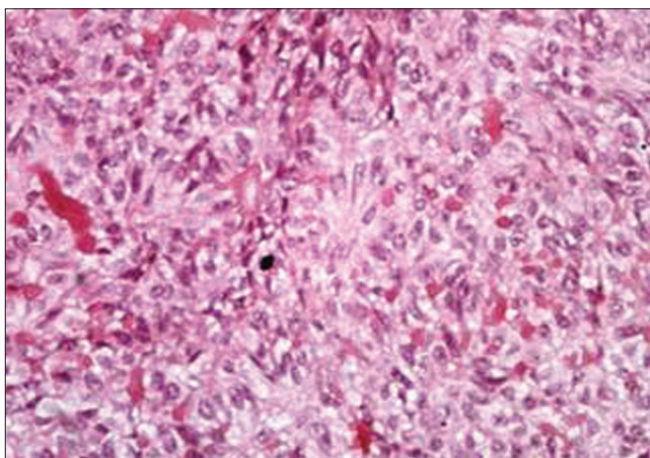


Figure 5: Cell-rich part of AOT showing eosinophilic tumor droplet material (×40)

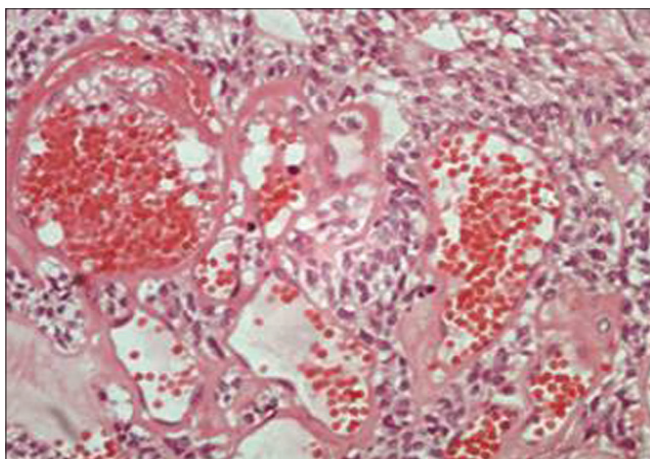


Figure 6: Thin-walled congested vessels characteristically showing marked degenerative (fibrinoid) changes of the endothelial lining (×40)

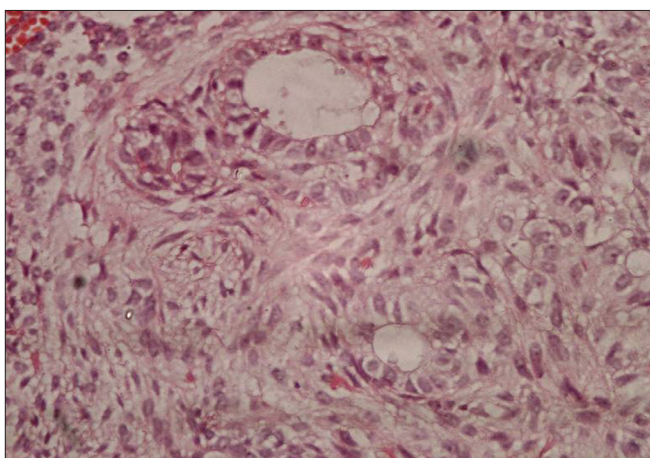


Figure 7: Characteristic tubular or duct-like structures lined by a single row of low columnar epithelial cells (×40)

well as root, but in our case, it was surrounding the radicular portion of the tooth.^[4] The cut section of the specimen shows extension of the tumor apically along the root past the

Cemento enamel junction, then distinguishing it from the more common dentigerous cyst.

Like all other odontogenic tumors, the specific stimulus that triggers proliferation of the progenitor cells of AOT is unknown. Because of its exclusive occurrence within the tooth bearing areas of jaws (most closely associated with an unerupted or impacted tooth) and its cytologic resemblance to the dental lamina and reduced enamel epithelium, there is an agreement that the AOT is of odontogenic origin.^[7,8]

WHO has described the histological features of the tumor as follows: “A tumor of odontogenic epithelium with duct like structures and with varying degree 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 histological appearance of all variants is identical and exhibits remarkable consistency. 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. Between the epithelial cells of the nodules and in the center of the rosette-like configuration is found eosinophilic amorphous material, often described as tumor deposits. Conspicuous within the cellular areas are structures of tubular or duct-like appearance. A third characteristic cellular pattern consists of nodules of polyhedral, eosinophilic epithelial cells with squamous appearance and exhibiting well-defined cell boundaries and prominent intracellular bridges. These islands may contain pools of amorphous amyloid-like material and globular masses of calcified material (thus the suggestion of a combination of calcifying epithelial odontogenic tumor and AOT). Another epithelial pattern has a trabecular or cribriform configuration. Occasional foci of mitotic activity can be traced. Induction of hyaline, dysplastic dentinoid material or calcified osteodentin has been described. Ultrastructurally, tumor epithelial cell types have been recognized, corresponding to the types that are evident on light microscopy.^[9] The connective tissue stroma is very loosely structured and contains thin-walled congested vessels characteristically showing marked degenerative (fibrinoid) changes of the endothelial lining, vessel wall, and perivascular connective tissue. It has been suggested recently that the tumor droplets represent some form of enamel matrix.

Immunohistochemical studies of the lesion suggest expression of keratin and vimentin in the tumor cells at the periphery of the ductal, tubular, or whorled structures. Amelogenin and enamelin in small mineralized foci are found in the tumor cells and in hyaline droplets. Since all variants show identical benign biological behavior and almost all are encapsulated, conservative surgical enucleation or curettage is the treatment of choice.^[10]

As all variants of AOT reveal an entirely benign biologic behavior and are well encapsulated, conservative surgical enucleation or curettage has proven to be the treatment modality of choice.^[11] In the present case enucleation was performed and no recurrence was seen after one year of follow up.

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