

Adenomatoid Odontogenic Tumor – A Case Report

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Abstract

Adenomatoid odontogenic tumor (AOT) a benign (hamartomatous) lesion of odontogenic origin is an uncommon tumor which affects young individuals with a female predominance. This lesion is most commonly located in the anterior maxilla and is usually associated with an impacted canine tooth. This report describes an AOT in the maxilla; illustrates the clinical, microscopic, and biological features of the tumor; and emphasizes the importance of the relationship between the dental follicle and the tumor tissue.

Keywords: Adenomatoid, Impacted tooth, Maxilla, Odontogenic tumor.

INTRODUCTION

An adenomatoid odontogenic tumor (AOT) was first described by Harbitz 1948 considered it a distinct entity, but it was classified by others as a variant of ameloblastoma.^[1,2] Philipsen and Birn proposed the name AOT in 1969 and regarded it not as a variant of ameloblastoma because of its clinical features and biological behavior indicate that it is a separate entity. Although there is evidence that the tumor cells are derived from enamel organ epithelium, investigators have also suggested that the lesion arises from remnants of dental lamina. This term was adopted by the World Health Organization (WHO) classification in 1971.^[3,4] Max and Stern coined the name adenomatoid odontogenic cyst in 2003. However, in 2005, the histologic typing of the WHO defined AOT as a tumor composed of odontogenic epithelium presenting a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma, and characterized by slow but progressive growth. This report describes an AOT in the maxilla; illustrates the clinical, microscopic, and biological features of the tumor; and emphasizes the importance of the relationship between the dental follicle and the tumor tissue.

CASE REPORT

A 14-year-old boy was referred to the Department of Oral and Maxillofacial Surgery, with swelling in the left maxillary anterior teeth region and good general health. Intraoral examination showed obliteration of the labial vestibule by expansion of the buccal cortical plate from the upper left canine and lateral incisor region. The permanent lateral incisor

was missing and an occlusal radiograph (Figure 1) revealed a well-circumscribed radiolucency around the impacted upper left lateral incisor. The lesion produced an expansion of tissue and extended into the alveolar process, disrupting the usual orientation of the anterior teeth. Based on the clinical and radiographic findings, the differential diagnosis was AOT, ameloblastic fibrous odontoma, clacifying odontogenic cyst, calcifying epithelial odontogenic tumor, infected dentigerous cyst, and unicystic ameloblastoma. The patient underwent surgery under local anesthesia. A mucoperiosteal flap was reflected in the left central incisor region to expose the labial aspect of the tumor (Figure 2). The labial cortex was very thin with several areas of complete resorption. The tumor was enucleated along with the impacted lateral incisor (Figure 3). The areas between the involved teeth were curetted well and the flap was sutured in place. Healing was satisfactory after the surgery. Histopathological examination revealed sheets of polygonal cells throughout the fibrous connective tissue stroma. The ductal lumina was surrounded by columnar epithelial cells and filled in some areas with eosinophilic material. In other places, amorphous calcified material was present (Figure 4). The histopathological report confirmed the diagnosis of an AOT.

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Figure 1: Occlusal radiograph

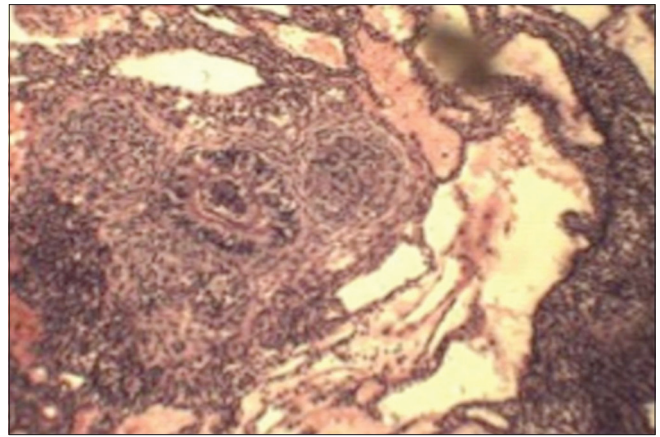


Figure 4: Histopathologic picture



Figure 2: Per-operative photograph



Figure 3: Excised tissue

DISCUSSION

AOT is a slowly growing lesion, with a predilection for the anterior maxilla (ratio of cases 2:1 relative to maxilla) of young males.^[5] 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 all variants is close to 2:1, the lesions are typically asymptomatic, but growth of the

types with central lesion results in cortical expansion, as in the case reported here.^[6] The involved teeth are commonly impacted and adjacent teeth may be slightly displaced. It can cause a painless hard swelling, as in the case reported here, and can be found on routine radiograph examinations. AOT, accounting for approximately 3% of all odontogenic tumors, is less frequent than odontoma, cementoma, myxoma, and ameloblastoma.^[7] It has been suggested that this tumor may be a hamartoma rather than a true neoplasm, but there is currently no evidence to resolve this dispute. 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 cyst does not envelop the roots. The origin of AOT is controversial. Some believe that they originate from the odontogenic epithelium of a dentigerous cyst. In addition to the anterior maxilla, the tumor has been reported in other areas of the jaw, such as the angle of the maxilla.^[1] Therefore, dental lamina remnants likely represent the progenitor cells for this benign odontogenic tumor. According to this hypothesis, the lesion grows while forming a cystic space next to or into a nearby dental follicle, leading to the “envelopmental theory.” In the case reported here, the lesion surrounded a fully form premolar, which suggests “envelopmental” pathogenesis. Recent reports indicate that the cells of an AOT usually differentiate toward an apparent ameloblastic phenotype but failed to achieve further functional maturation. The 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.”^[8] The histologic 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 nest 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.^[9,10] This same pattern was observed in our case. 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. 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. Amelogenin and enamelin in small mineralized foci are found in the tumor cells and in hyaline droplets.^[6] Since all variants show identical benign biological behavior and almost all are encapsulated, conservative surgical enucleation or curettage is the treatment of choice. Recurrence has been reported has been reported in very few cases.^[1]

CONCLUSION

The present case is a rare report of an AOT presented in the maxilla of a 14-year-old boy involving a permanent lateral

incisor and showing hybrid tumor-like histopathological pictures and its subsequent management.

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