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Case Report

Aggressive adenomatoid odontogenic tumor of mandible showing root resorption: A histological case report

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is a benign odontogenic tumor with slow but progressive growth. The three variants: Follicular, extra follicular (both central type), and peripheral present with identical histologic findings. This case report describes a patient with a large AOT in the mandible of the extra follicular type which is the less common of the two central types. It also strikes as an unusual case as it shows significant root resorption of the involved displaced teeth which is not generally reported in AOT's.

Key Words: Adenoameloblastoma, adenomatoid, hamartoma, root resorption

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a benign non-neoplastic hamartomatous lesion originating from dental lamina or its remnants. The lesion is known by many names, including adenoameloblastoma, adenoameloblasticodontoma, epithelial tumor associated with developmental cysts, ameloblastic adenomatoid tumor, and adenomatoid or pseudoadenomatous ameloblastoma.[1-3] Due to its non-invasive harmless nature, Philipsen and Birn in 1969 introduced the term "AOT" which was adopted by WHO in 1971.[4] Based on the current knowledge, three main types of AOTs are known, namely, (1) follicular (or pericoronal), (2) extrafollicular (or extracoronal), and (3) peripheral (or extraosseous/gingival). In a recent retrospective study,[5] 70.8% were of follicular type of which almost half the number were females. The extrafollicular variety accounted for 26.9% again with a M: F ratio of (close to) 1:2. The rare peripheral variant (2.3%) showed a remarkable M:F ratio of 1:6.3.

The histogenesis of AOT remains controversial. Some authors suggest it to be a true benign, non-aggressive, non-invasive neoplasm and others conceptualize it as a developmental hamartomatous odontogenic growth. Here, we report a case in which AOT shows an unusual, aggressive behavior suggesting it to be a true benign but an aggressive neoplasm.

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CASE REPORT

A 17-year-old-male patient reported to the Department of Dental Sciences with a gross asymmetry over the right anterior region of the mandible. The patient gave a history of loosening of teeth since 1 year. Intra-oral examination revealed a bony hard swelling in the right mandibular region extending from central incisor to first molar [Figure 1]. Right mandibular lateral incisor, canine, first premolar, and second premolar were mobile and gave no response on electrical pulp testing. On palpation the swelling was non-tender, irregular in shape, and measuring 2 cm × 3 cm. Orthopantomogram showed a large well-circumscribed radiolucency extending from right mandibular central incisor to the mesial root of first molar [Figure 2].

Due to the expansive growth there was deviation of the roots of right mandibular canine and first premolar. Root resorption was also evident in relation to the first premolar, second premolar, and mesial root of first molar on the right side. Based on these features, a differential diagnosis of odontogenic cysts and tumors (like odontogenic keratocyst, lateral dentigerous cyst, AOT, calcifying epithelial odontogenic tumor) was given. Other differential diagnosis included central giant cell lesions, lateral periodontal cyst, lateral radicular cyst, and the much rarer central benign mesenchymal neoplasms. The lesion was operated under local anesthesia and the specimen was sent for histopathologic examination. The mobile teeth, that is, mandibular lateral incisor, canine, premolars, and the first molar were removed along with the lesion [Figure 3]. Grossly, the lesion was white to tan, nodular tissue with cystic spaces containing yellowish brown semisolid material [Figure 4]. Microscopic examination revealed extremely vascular encapsulated lesion showing multivariate patterns of cellular arrangements ranging from sheets of polygonal cells arranged in ductal pattern, rossetes to solid sheets of cells [Figure 5]. In the center of these ducts, eosinophilic amyloid-like material was also seen. The solid lobular masses showed numerous spindle to columnar hyperchromatic cells with interspersed deposits of eosinophilic hyaline-like material. Mitotic figures were also seen in the lobules [Figures 6 and 7]. The above features were consistent with the diagnosis of AOT.

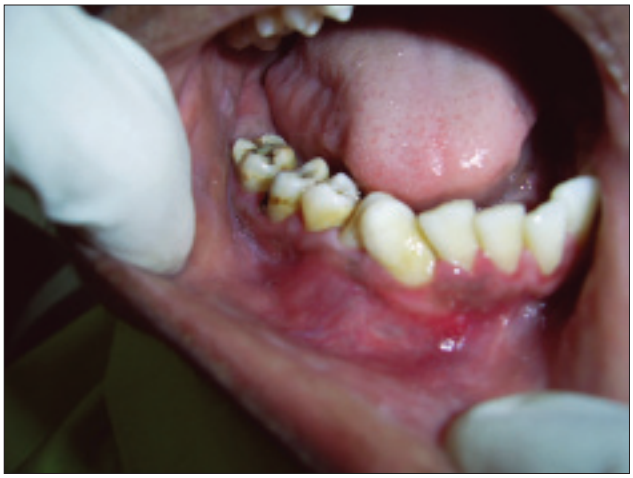

Figure 1: Intra-oral picture showing swelling in the right mandibular buccal sulcus

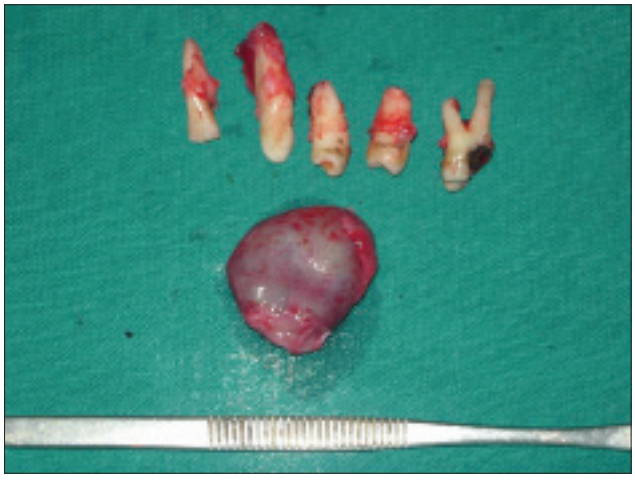

Figure 3: Excised tumor specimen along with extracted teeth

DISCUSSION

AOT is a slow-growing tumor with a higher rate of occurrence in the anterior maxilla of young females. It can occur between 3 and 82 years of age but the majority (68.6%) occurs in the second decade of life, making this a unique feature among odontogenic tumors. AOT presents as three clinical variants: Follicular, extrafollicular, and peripheral.[6-8]

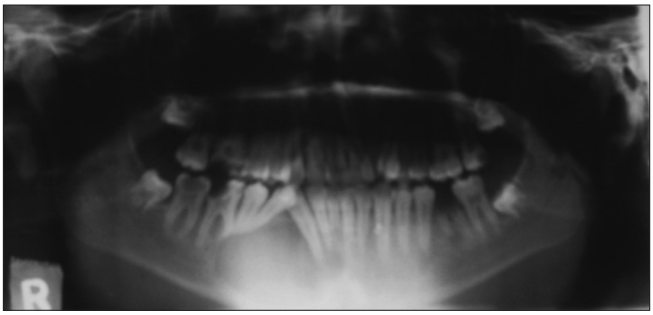

Figure 2: Orthopantomogram showing well-defined radiolucency in the right mandibular body with tooth displacement and root resorption of 43, 44, 45, and 46

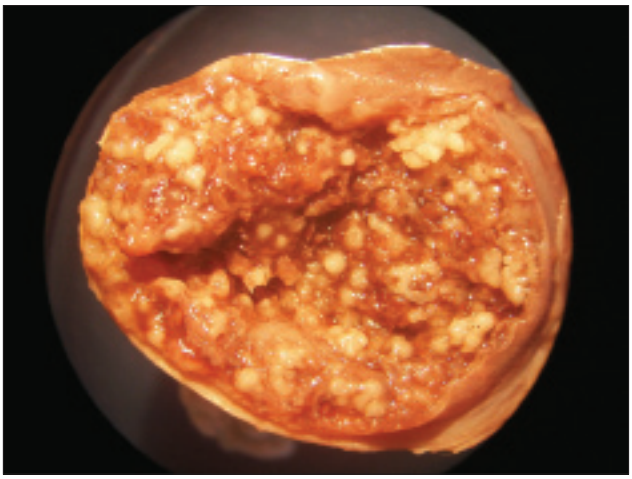

Figure 4: Cut surface of the gross specimen showing white to tan, nodular tissue with cystic spaces

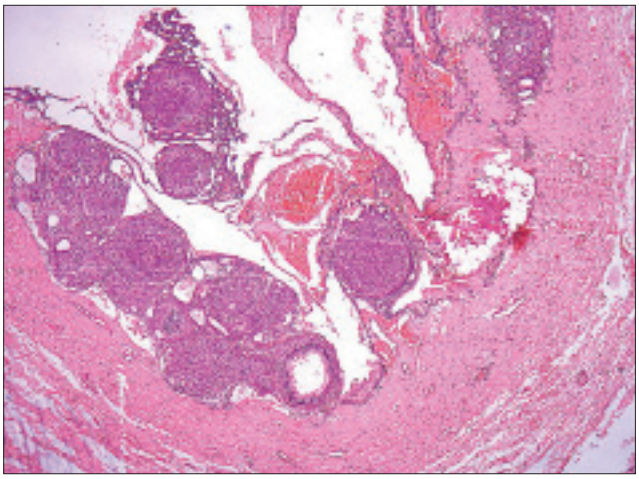

Figure 5: Photomicrograph showing the typical ductal and rosette pattern of tumor cells in an adenomatoid odontogenic tumor

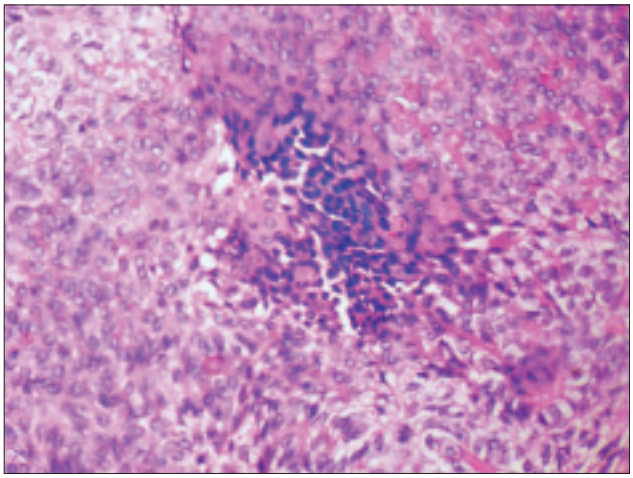

Figure 6: Photomicrograph showing solid lobular masses with numerous spindle to columnar hyperchromatic cells

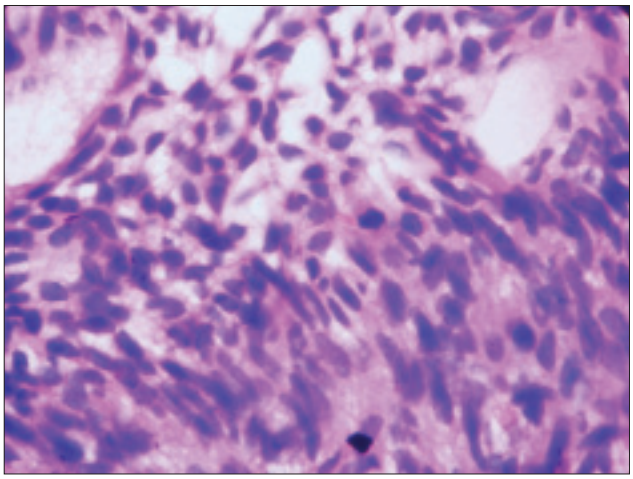

Figure 7: Photomicrograph showing spindle-shaped hyperchromatic cells with pleomorphism and presence of mitotic figures

Of these, the follicular and extrafollicular are the central variants and account for 96% of the lesions of which 71% are the follicular type.[9] The two central variants are found more commonly in the maxilla than the mandible in a ratio of 2.1:1. The case reported here is uncommon as it is present in the mandible as the extrafollicular type. AOTs are most commonly diagnosed on radiographic examination as they are usually small asymptomatic lesions. Rarely, if the lesion is large it can cause a painless hard swelling of the involved jaw bone, as in the case reported here. The most common radiographic appearance is that of the follicular type seen as a radiolucency surrounding the crown of an unerupted tooth. Nearly, 30% of the central AOTs (extra follicular) are not pericoronal and demonstrate a relationship to the roots of adjacent or nearby teeth that ranges from lateral or interproximal to periapical to no relationship at all. Divergence of roots and displacement of teeth occurs more frequently than root resorption.[9] In this case, all the permanent teeth were erupted and the radiolucency was seen between and apical to multiple teeth in the region causing their displacement and root resorption.

WHO has defined the histologic features of AOT as "A tumor of odontogenic epithelium with duct-like structures and with varying degree of inductive change 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 picture of all three types is similar. The tumor is made up of a multi-nodular proliferation of spindle, cuboidal, and columnar cells in a variety of patterns comprising of scattered duct-like structures, eosinophillic material, and calcifications in several forms; delimited by a fibrous capsule of varying thickness. Cytologic atypia is never a prominent feature. In the present case, numerous hyperchromatic cells and mitotic figures were seen which are not usual features of AOT. The duct-like structures with varying lumen size are lined by a single layer of cuboidal to columnar epithelial cells that have nuclei that are frequently polarized away from the lumen.[10] 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, eosinophilic amorphous material is found, often described as tumor deposits. Conspicuous within the cellular areas are structures of tubular or duct-like appearance.[9]

During the last few years, several studies have been published dealing with the immunohistologic properties of AOT. Immunohistochemically, the classical AOT phenotype is characterized by a cytokeratin (CK) profile similar to follicular cyst and/or oral or gingival epithelium based on positive staining with CK5, CK17, and CK19.[11] On the other hand, the classical AOT is negative for CK4, 10, 13, and 18. Recently, Crivelini, *et al*.[12] detected the expression of CK14 in AOT and concluded that this probably indicate its origin in the reduced dental epithelium which is also positive for staining with CK14 antibodies. Positive reactions for amelogenin in limited areas in AOT are also reported as well as in ameloblasts and in the immature enamel matrix.[13] Recent immunohistochemical studies show that all cellular types that composed of AOT showed nuclear positivity for p63 indicating a basal characterization in the different cellular components. According to its benign character and low potential for recurrence, AOT revealed a scant proliferative activity (2-3% nuclei showed Ki-67 positivity) limited to some epithelial nodules of fusiform appearance.[14] In cases where unusual behavior-root resorption as in our case is seen, a detailed immunohistochemistry should be done for better understanding of the nature of the tumor. Due to lack of availability of the facility, it could not be performed for this case but the unusual radiologic and histopathologic features and its rare location need to be added to the literature for reference in future cases.

Conservative surgical enucleation is the treatment modality of choice. Recurrence of AOT is exceptionally rare. Only few cases have been reported in which the recurrence of this tumor occurred. Therefore, the prognosis is excellent. But in cases where unusual findings are seen, the follow up should be for a longer period to check for recurrences, as seen in the present case.

CONCLUSION

The authors conclude that the case presented here is a rare and aggressive AOT as it is the extrafollicular variant located in the mandible causing root resorption and associated with increased mitosis.

Hence it should be distinguished from the other more common odontogenic lesions.

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