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Case Report

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

Ramandeep Saluja[1], Gurkiran Kaur[2], Preetinder Singh[3]

[1]Departments of Oral Pathology and Microbiology, [3]Department of Periodontology and Oral Implantology, Swami Devi Dyal Hospital and Dental College, Barwala, District Panchkula, Haryana, [2]Department of Oral Pathology and Microbiology, Gian Sagar Dental College and Hospital, Ramnagar, Rajpura, District Patiala, Punjab, India

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Address for correspondence:
Dr. Ramandeep Saluja,
House No 1638, PHASE 10,
District Mohali, Punjab,
India.
E-mail: doc raman_7@hotmail.com

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]

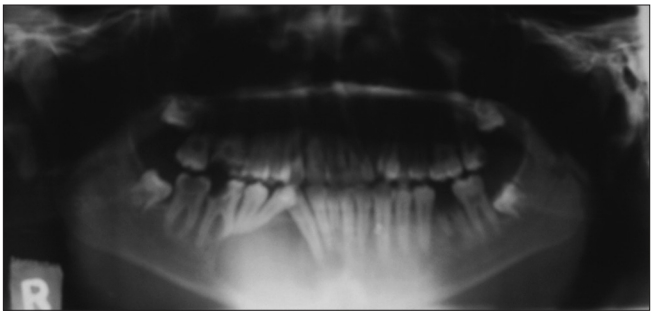

Figure 2: Orthopantomogram showing well-defined radiolucency in the right mandibular body with tooth displacement and root resorption of 43, 44, 45, and 46

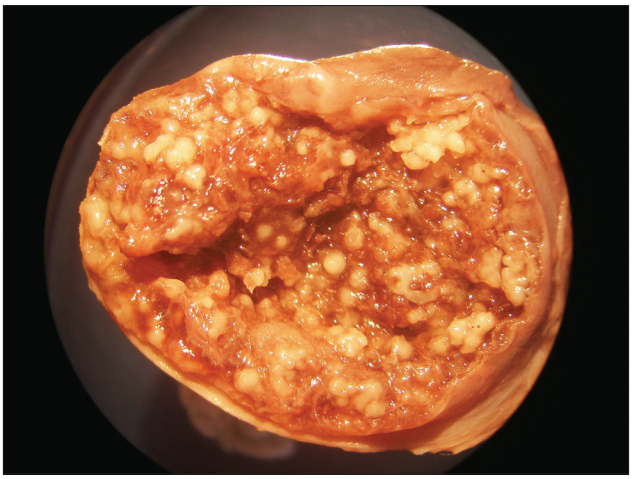

Figure 4: Cut surface of the gross specimen showing white to tan, nodular tissue with cystic spaces