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Case Report

ADENOMATOID ODONTOGENIC TUMOR ARISING FROM THE MANDIBULAR MOLAR REGION: A CASE REPORT AND REVIEW OF THE LITERATURE

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Abstract

This case report describes a rare case of an adenomatoid odontogenic tumor (AOT) arising in the mandibular third molar region of a 20-year-old female. The tumor was a well-circumscribed intraosseous lesion with an embedded tooth. Histologically, the induction of extensive globular calcification was evident in addition to the characteristic histological features of AOT. The present case lends support to the categorization of AOT as an odontogenic tumor consisting of a disorderly mixture of odontogenic epithelium and odontogenic ectomesenchyme with calcification.

Key words: Adenomatoid odontogenic tumor—AOT—Odontogenic tumor—Mandibular

INTRODUCTION

Adenomatoid odontogenic tumors (AOT) are rare, slow growing, benign, odontogenic, epithelial tumors which usually arise in the second or third decade. AOT are usually located in the anterior region of the maxilla; they normally, produce slow swelling without pain. The tumor growth may cause displacement of teeth rather than root resorption. AOTs comprise only 0.1% of tumors and cysts of the jaw. Philipsen et al. reported that they account for 3 to 7% of all odontogenic tumors. In Philipsen’s report, studies in Nigeria (Sawyer/Ajagbe et al.) showed a higher frequency, but other studies (Regezi et al. in the USA and Happonen et al. in Finland, among others) record a lower frequency. We herein report a rare case of AOT and review the literature.

CASE REPORT

A 20-year-old female with a painless swelling of the mandibular third molar area was
admitted to the Tokyo Dental College Department of Oral Surgery. Clinical examination revealed a circumscribed, bucco-lingual swelling. Radiographic examination showed a unilocular radiolucency extending with an embedded tooth (Fig. 1). No root resorption or displacement of neighboring teeth was seen. The lesion was completely enucleated under the clinical diagnosis of a dentigerous cyst, and sent to the Department of Clinical Pathophysiology of the Tokyo Dental College for pathological diagnosis.

RADIOLOGIC FINDINGS

A radiolucent lesion with a regular border was seen in the mandibular third molar area. The radiolucency associated with the unilocular lesion extended to the apical regions of the unerupted teeth. No displacement of neighboring teeth due to tumor expansion or root resorption was seen (Fig. 1).

HISTOPATHOLOGICAL FINDINGS

The lesion was a well-defined mass 22 × 18 mm in size that was surrounded by a thick, fibrous capsule. The mandibular third molar was encapsulated. After the specimen was fixed with 10% formalin, paraffin sections were prepared for light microscopy and stained with hematoxylin and eosin using routine methods. Histological examination revealed amorphous eosinophilic droplets and foci of dystrophic calcification scattered in and around the epithelial elements (Fig. 2). The epithelial elements consisted of polyhedral cells in loose and irregular arrangements, spindle cells, and tall columnar cells that formed duct-like structures as a result of the degeneration of the stromal tissue. The nuclei of the columnar cells were polarized away from the central space. The interstitium consisted of loosely arranged fibrous connective tissue containing dilated vessels, and a lot of small cystic spaces were present.

Fig. 1 Preoperative panoramic radiography showing a well circumscribed unilocular radiolucency around the third molar (arrowhead)

Fig. 2
a. Dysplastic calcification H-E staining
b. Duct-like and adenoid structure
Arrowhead: Tall columnar cell
DISCUSSION

AOT is defined as a tumor of the odontogenic epithelium with duct-like structures and varying inductive changes in the connective tissue\(^{3-5,19}\). Preoperatively, AOTs have been diagnosed as various types of disease, because of the variety of clinical and radiographic features of the lesions. Radiographically, they frequently look like a dentigerous cysts or follicular cysts. The radiolucency associated with an adenomatoid odontogenic tumor may extend more apically than that of a dentigerous cyst\(^5\).

The tumors are usually 1.5 to 3 cm\(^5\), but larger lesions have been reported in the literature\(^9,21\). The lesions usually appear unilocular radiographically, but at least five cases of multilocular appearance have been reported\(^5\). Larsson reported a case of multiple AOT-like jawbone lesions in a young patient\(^9\). AOT lesions may often appear completely radiolucent; however, they contain fine calcifications\(^7,21\), a feature that may be helpful in differentiating AOT from dentigerous cysts.

The calcified materials seen in AOT have been considered to be a form of enamel\(^8\), dentin\(^22\), enamel and dentin\(^11\), cementum\(^18\), dentin and cementum\(^12\) and dystrophic calcification\(^7\), but their exact nature remains controversial\(^10\). In the present case, no dental hard tissue formation was observed; only dystrophic calcification was seen.

Ishikawa and Mori suggested that most of the duct-like structures are formed by degeneration of the stromal tissue, because some of the luminal content had the same staining characteristics as the mesenchymal fibrous tissue\(^7\). A direct transition between the duct-like lumen and the degenerating stroma was clearly evident. The characteristic features of this tumor are probably due to both the highly differentiating ability of the epithelial component and the lack of mesenchymal tissue receptive to the influence of the epithelium.

Information on the incidence and prevalence of individual odontogenic tumors is still not available. However, AOT is a particularly rare tumor that represents 3% to 7% of all odontogenic tumors\(^2,5,8,14,15,21\). In our laboratory, AOT was diagnosed in 9 cases during the period from 1967 to 2004 (Table 1), which comprises only 1.8% of odontogenic tumors (10/559) seen during that time.

The age of patients with AOT ranges between 3 and 82 years of age at the time of diagnosis\(^14,15\), although those tumors are uncommon in patients older than 30. The age distribution peaks in the second
decade; two-thirds of AOT cases are diagnosed in the second decade. In our laboratory, only two cases have been older than 30. In Philipsen’s report, the ratio of patients at least 30 years old is 7.5% (25/32). Among Japanese cases, 10.5% of the patients are reported to be over 30 years old.

The maxilla is involved nearly twice as frequently as the mandible, and unerupted permanent teeth are associated with this lesion in one-third of the cases. Three quarters of these lesions involve the anterior aspect of the jaw, and the canine area of the maxilla is the most common site of development. In our laboratory, 3 cases of AOT were in the maxilla region and 6 cases in the mandible. It is remarkable that the mandible is more frequently involved than the maxilla and that 2 cases were found in the molar region of the mandible. All these cases were of the intraosseous type.

AOT affects females more than males at almost a two to one ratio, according to the published English literature. AOT-cases in Asians show a male:female ratio of 1:2.3, whereas cases outside of Asia show a ratio of 1:1.4, a difference that is statistically significant. In Japan, these lesions are found three times more frequently in females than in males, which can not be explained at present. In our laboratory, 2 cases were male and 7 cases were female (Table 1).

Enucleation and curettage is the most common treatment modality for this tumor, and recurrence is extremely rare. Only three cases of recurrent AOT among 750 cases have been reported, and all the recurrent cases were in the Japanese literature. Careful follow-up examinations should be conducted in this disease.

REFERENCES


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