

Adenomatoid Odontogenic Tumor of the Mandible

Yadavalli Guruprasad¹, Prashanth Ramesh Prabhu²

Abstract

Adenomatoid odontogenic tumor (AOT) is an uncommon benign hamartomatous lesion of odontogenic origin, which affects young individuals, with a female predilection and mainly occurs in the second decade. It is located more often in the maxilla and in most cases associated with an unerupted permanent tooth. It is considered as a hamartoma because of its limited size, minimal growth potential, and lack of recurrence in most cases. AOT accounts for about 1% until 9% of all odontogenic tumors. We present a rare case of an AOT of mandible in a 34-year-old female patient.

Key words: Adenomatoid odontogenic tumor, mandible.

Introduction

Adenomatoid odontogenic tumor (AOT), an uncommon benign epithelial lesion of odontogenic origin, was first described by Dreibaldt in 1907 as a Pseudoameloameloblastoma.¹ However, a variety of terms have been used to describe this tumor. Unal et al., produced a list containing all nomenclatures for AOT reported in the literatures.² Many different names like adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum or teratomatous odontoma, have been used before to define the lesion currently called AOT. In 1999 Philipson and Reichart presented a review based on reports published until 1997 which showed some interesting aspects regarding epidemiological figures of this tumor.³ Since then numerous case reports of AOT have been published. From the early 1990s onwards 65 single cases of AOT (excluding case series of more than 10 cases) have been published. The mean age was 13.2 years (range 3 until 28 years) and the female: male ratio was 2.3:1. The AOT was predominantly found in the upper jaw (maxilla: mandible = 2.6:1). As per the various case series published in the literature⁴ and comparing these data with the single case reports, AOT has a prevalence of odontogenic tumors between 1.2% in Caucasian and 9% in black African patients.⁵ The tumor is most often diagnosed in the second decade of life and women are affected twice as frequently as men. The AOT is over two times more located in the maxilla than in the mandible and the anterior jaw is much more affected than the posterior area. According to Philipson and Reichart the AOT appears in three clinicotopographic variants: follicular, extra follicular and peripheral.⁵ The follicular and extra follicular variants are both intrabony and account for approximately 96% of all AOTs of which 71% are of follicular type.

Case report

A 34-year-old female patient was referred to the Department of Oral and Maxillofacial Surgery for the evaluation of an asymptomatic swelling in the right lower jaw since six months. The lesion had been slowly increasing in size since it was first noticed. There was no history of trauma, pain, parasthesia or lymphadenopathy. On clinical examination,

the lesion extended from right canine to first molar on the same side intraorally and was soft in consistency on palpation (Figure 1,2). A panoramic radiograph showed a well-circumscribed radiolucency in relation to the lower right premolars (Figure 3). Enucleation and curettage was done under local anaesthesia using intraoral approach (Figure 4,5). Histopathology revealed epithelium in the form of whorled masses of spindle cells as well as sheets and plexiform strands. Rings of columnar cells give rise to duct-like appearance (Figure 6). Calcification is sometimes seen and may be extensive. Clinical and radiographic follow-up examination was performed six months after surgery. There was no evidence of recurrence and no apical resorption of the adjacent teeth could be observed (Figure 7,8). With respect to the age of the patient and the localization of the AOT in the lower jaw, the reported case is a rare example of this tumor entity. Beyond it our case supports the above mentioned general description of AOTs.

Discussion

Adenomatoid odontogenic tumor is a slow growing lesion, constituting only 3% of all odontogenic tumors with a predilection for the anterior maxilla (ratio 2:1) relative to mandible and is usually associated with impacted canine, of young females in the second decade of life.⁶ In our case the lesion occurred in the posterior mandible which is unusual. Clinical features generally focus on complaints regarding a missing tooth. The lesion usually present as an asymptomatic swelling which is slowly growing and often associated with an unerupted tooth. However, the rare peripheral variant occurs primarily in the gingival tissue of tooth-bearing areas.⁷ Unerupted permanent canine are the teeth most often involved in AOTs. The radiographic findings of AOT frequently resemble other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, ameloblastomas, odontogenic keratocysts and periapical disease.^{7,8} Whereas the follicular variant shows a well-circumscribed unilocular radiolucency associated with the crown and often part of the root of an unerupted tooth. The radiolucency of the extra follicular type is located between, above or superimposed upon the roots of erupted perma-

¹Asst. Professor, Dept. of Oral and Maxillofacial Surgery, AME'S Dental College Hospital & Research Centre, Raichur, ²Asst. Professor, Department of Oral and Maxillofacial Pathology, Sinhgad Dental College & Hospital, Pune, India.
Correspondence: Dr. Yadavalli Guruprasad, email: guru_omfs@yahoo.com



Figure 1: Photograph showing frontal view of the patient



Figure 2: Clinical appearance of the swelling on the right side intraorally

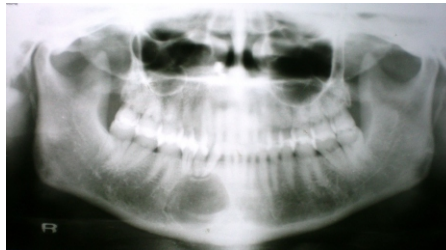


Figure 3: Panoramic radiograph showing unicystic radiolucent lesion in the lower right jaw in relation to premolars with a clear demarcation



Figure 4: Intraoperative photograph showing the lesion which is exposed using crevicular incision



Figure 5: Intraoperative photograph after complete excision

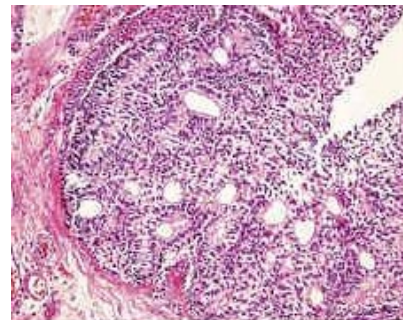


Figure 6: Tumor with fibrous connective tissue capsule. Nodular aggregates of cells and duct-like structures are seen (H and E, X 40)

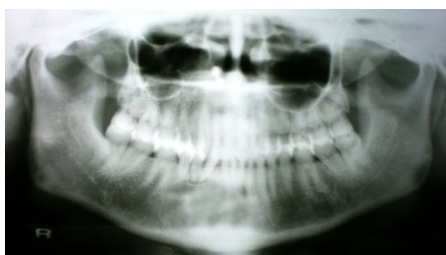


Figure 7: Postoperative panoramic radiograph after six months



Figure 8: Postoperative clinical photograph after six months

ment teeth. Displacement of neighboring teeth due to tumor expansion is much more common than root resorption. The peripheral lesions may show some erosions of the adjacent cortical bone.⁸ Comparing diagnostic accuracy between intraoral periapical and panoramic radiographs, it was found that intraoral periapical radiographs allow perception of the radiopacity in AOT as discrete foci having a flocculent pattern within the radiolucency even with

minimal calcified deposits while panoramic often do not.^{7,8} Those calcified deposits are seen in approximately 78% of AOT. In addition, in one recently reported case MRI was found useful to distinguish AOT from other lesions, even if it is difficult to distinguish on periapical ordinary radiographs.^{8,9} Conservative surgical enucleation is the treatment modality of choice. For periodontal intrabony defects caused by AOT, guided tissue regeneration with membrane

technique is suggested after complete removal of the tumor.^{9,10} Recurrence of AOT is exceptionally rare. Therefore, the prognosis is excellent.¹⁰

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