Case Report

Extrafollicular Adenomatoid Odontogenic Tumor of the Maxilla: A Case Report

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Abstract

Adenomatoid odontogenic tumor (AOT) is an uncommon benign odontogenic tumor representing approximately 3% of all odontogenic tumors. In about 74% of cases, this tumor is associated with an impacted tooth, of which two-third of the cases is in relation to the maxillary anterior tooth. Rarely, this lesion can also occur in the absence of an impacted tooth within jaw bones. These variants of AOT are referred as extra follicular AOT. As the clinical picture and radiographic picture varies from the conventional AOT, the diagnosis is often challenging. Here, we report a case of extra follicular AOT of the anterior maxilla in a 22-year-old female patient.

Keywords: Adenomatoid Tumor; Maxilla; Odontogenic Tumors.

Introduction

Adenomatoid odontogenic tumor (AOT) is a benign, uncommon, slow growing tumor representing 3% of all odontogenic tumors [1,2]. The tumor is usually associated with impacted tooth, frequently maxillary canines. It often causes expansion of surrounding bone and the displacement of adjacent teeth. However, owing to the slow growing nature, the lesion remains unnoticed until obvious deformity is developed [3]. A rare intraosseous variant of AOT which is not associated with an unerupted or impacted tooth is reported. This entity is referred as extra follicular AOT [4]. In this case report, we describe an extra follicular AOT which presented as a swelling in the maxillary anterior region of a 22-year-old female patient.

Case Report

A 22-year-old female patient was referred to the Department of Oral Medicine and Radiology, Faculty of Dentistry, Annamalai University, by a Private Dental Practitioner for the evaluation of painless swelling in the maxillary anterior region. She reported the swelling was present since 5 years, and it gradually increased in size to attain the present size. There was no history of trauma, pain, discharge, or any other symptoms related to the lesion. The patient was apparently healthy, and her vital signs were within normal limits. On extra oral examination of the head and neck, chronic lymphadenopathy was seen on the right and left submandibular lymph nodes. Facial asymmetry was observed due to the swelling in relation to left nasolabial fold region. Intraorally, a single, well-defined swelling measuring 3 cm × 2 cm was observed on the alveolus between the left maxillary lateral incisor and canine (Figure 1). A mild labiopalatal expansion was seen on the maxillary alveolus with obliteration of labial vestibule. Surface of the swelling was smooth with normal overlying mucosa. There was no pulsation visible or palpable. The swelling was firm to hard in consistency and nontender on palpation. Fine needle aspiration yielded no fluid. Periapical and panoramic radiographs showed circumscribed radiolucent area with fine calcifications between maxillary left lateral incisor and canine. Displacement of these teeth was seen but with no root resorption. Occlusal radiograph presented a moderate labiolingual expansion, and a thinned

Figure 1: Well defined swelling on the alveolus between left maxillary lateral incisor and canine

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labial cortical plate (Figure 2). Medical Computed tomography (CT) scans demonstrated well-demarcated hypodense lesion with scattered hyperdense foci, and loss of cortical bone (Figure 3). The possibilities of ossifying fibroma, adenomatoid odontogenic tumor (AOT), ameloblastoma, and keratocystic odontogenic tumor were considered in the differential diagnosis preoperatively. The lesion was totally enucleated and removed, and the tissue was submitted to histopathological examination.

Microscopy of the specimen revealed whorls of epithelial Islands surrounded by a well-defined mature capsule. The cells of epithelial Islands showed round to oval nuclei (Figure 4). Many well-formed pseudoducts lined by tall columnar cells were evident. The lumen showed eosinophilic material and typical rosette form was also evident in many areas. Abundant calcifications were also seen, especially in the periphery. Based on these features, the final diagnosis of AOT was obtained. Healing was uneventful after surgical enucleation of the lesion (Figure 5).

**Discussion**

AOT is a benign odontogenic tumor of epithelial origin thought to originate from remnants of a dental lamina or enamel organ. The lesion was first described by Drebladt in 1907 [1]. Philipsen and Birn proposed the name AOT in 1969 and differentiated it from ameloblastoma [2]. Most of the cases are reported in the second decade of life with a female predilection of 2.3:1 and site predominance for anterior maxilla associated with impacted maxillary canine [3]. The age, sex, and the location of our case were consistent with the literature. The patient with AOT usually reports with a complaint of slow growing asymptomatic hard swelling. Generally AOT does not exceed 1–3 cm in diameter. It causes cortical expansion, impaction, and noneruption of the involved tooth and displacement of the adjacent teeth. Thus, majority of the AOT cases are associated with unerupted/impacted tooth. Rarely the affected tooth manages to erupt through the lesion as in our
case [3]. Exact nature of pathogenesis of AOT is still unclear and remains controversial. Some authors believe it to be a hamartoma of the remnant odontogenic epithelium [4].

Philipsen and Reichart considered AOT to be a non-invasive slow-growing benign lesion (hamartomatous) [5]. However, a study conducted in Japan by Takahashi et al. detected transferrin by immunohistochemistry in part of the tumoral cells and suggested neoplastic nature of AOT [6].

The WHO histological typing of odontogenic tumors, jaw cyst, and allied lesions (2005) has defined AOT as a tumor of odontogenic epithelium with duct-like structures and with varying degree of inductive changes in the connective tissue [7].

The tumor has three clinicopathologic variants, namely intraosseous follicular, which is associated with an unerupted tooth (74%), whereas intraosseous extrafollicular type (24%) which has no relation to an impacted tooth as in the case presented here, and the peripheral variant (3%) which is attached to the gingival structures [8].

Radiographically the follicular type appears as well circumscribed pericoronal radiolucency with or without radiopacities associated with an unerupted tooth. At an earlier stage, it appears to be nothing more than an enlarged dental follicle radiographically [3]. Reichart and Philipsen subdivided extrafollicular AOT into four subtypes (E1–E4) based on radiographic appearance [9].

A lesion falling in E1 subtype has no relation to tooth structures either erupted or unerupted and is extremely rare. In any process of eruption of associated tooth through the lesion, spatial relationship between AOT in relation to the tooth can be on the lateral root surface (E2; inter-radicular), at a root apex (E3; radicular/periapical or superimposed over the root (E4). The case which we reported here fits into the E2 subtype of extrafollicular AOT. In two-third of the cases of AOT, the lesion may contain discrete foci and a flocculent pattern of scattered radiopacities which are best seen in intraoral periapical radiography. Root resorption of adjacent teeth is rare [3]. The radiographic differential diagnosis would include dentigerous cyst, calcifying epithelial odontogenic tumor, and odontogenic keratocyst [3]. While dentigerous cyst encircle only the crown of a tooth, the AOT often appears to envelop the crown as well as the root and may contain fine radiopaque calcifications. An extrafollicular AOT in the maxillary anterior region may mimic a fissural cyst, odonogenic tumors such as ameloblastoma, keratocystic odontogenic tumor, and fibrousosseous lesion like ossifying fibroma [9].

Unlike follicular variant of AOT, the pathogenesis of the extrafollicular variant remains unclear. However, the available reviews in the literature suggest that some extrafollicular AOTs may arise as a secondary occurrence within pre-existing odontogenic cysts or cystic tumors. An unusual subvariant of the extrafollicular type of AOT may radiographically imitate periapical diseases, which is initially doubtful of small periapical pathology [10, 11].

The histological findings for AOT are characterized by odontogenic epithelium with duct-like structures and with varying degree of inductive changes in the connective tissue [9]. The tumor may contain cystic areas and in some cases the solid lesion may be confined only to the walls of cystic spaces. The tumor may contain pools of amyloid-like material and globular masses of calcified material [9]. The tumor is usually well-encapsulated and shows typical benign behaviour [3].

Therefore, conservative surgical enucleation produces excellent outcome without recurrence. Our patient has been under follow-up for 12 months with no evidence of recurrence.

References