Adenomatoid Odontogenic Tumor: An Uncommon Mandibular Case Report

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ABSTRACT

The adenomatoid odontogenic tumor is a hamartomatous benign neoplasia of odontogenic origin. It appears mostly in young patients and females, the maxillary region being the most affected. It is a slow-growing, asymptomatic lesion. It is related to non-erupted teeth, mainly canines. Lesions of this type can be clinically classified as a follicular, extrafollicular and peripheral lesions. The treatment for these lesions is enucleation and curettage of affected area. Here we report a case of the adenomatoid odontogenic tumor (AOT) in the mandibular right region of a young man aged 20 years, and no recurrence has been observed after one-month follow-up.

KEYWORDS: Adenomatoid, Odontogenic Tumor, Mandibular, Uncommon, Benign Neoplasia

INTRODUCTION

The adenomatoid odontogenic tumor was first described in 1907 by Dreibladt, as a pseudo adenoameloblastoma.1 Over years, a variety of terminologies has been used to designate this extremely fascinating entity like adenoameloblastoma, adenoameloblastic odontoma, epithelial tumor associated with an ameloblastic adenomatoid tumor, developmental cysts, and adenomatoid or pseudo adenomatous ameloblastoma. Philipsen and Ben suggested the name of adenomatoid odontogenic tumor in 1969 and that it won’t be considered as a variant of ameloblastoma because of its different behaviour.1,2

The adenomatoid odontogenic tumor is also called «A two-thirds tumor », because 2/3rd of the adenomatoid tumors occur in young females, 2/3rd of the adenomatoid tumors occur in the maxilla, 2/3rd of the cases of these tumors are associated with unerupted teeth, and two-thirds of the affected teeth are canines.3

There are 3 variants of adenomatoid odontogenic tumor. The follicular type (accounting for 73% of cases), which has a central lesion associated with an embedded tooth, the extrafollicular type (24% of case), which has a central lesion and no connection with the tooth, and the peripheral variety (accounting for 3% of cases). The aim of this paper is to describe a case of the adenomatoid odontogenic tumor (AOT) in the mandibular right region of a young man aged 20 years, and no recurrence has been observed after one-month follow-up.

CASE REPORT

A 20-year-old young male reported to the Oral Surgery Department complaining from a swelling in the right inferior front tooth region evolving since 5 months. The medical history of the lesion revealed that initially the swelling was small in size, and gradually it increased to reach up to the present size. It was not associated with any pain or discharge, with no history of trauma.

The Intraoral examination revealed a distinct swelling in the anterior region of the mandible from the 45 to 33. The overlying mucosa was normal, and there was no paresthesia in the mandibular region. Because of the swelling, the central and lateral mandibular incisors were deviated to the right, and the patient had retained his right canine (Figure 1).

Fig 1 : Intraoral examination revealing the Presence Of a mandibular Swelling extended from the 45 to 41 with a right deviation of the lateral mandibular incisors

Panoramic radiograph and computed tomography incidences were performed, and which revealed a well defined unilocular radiolucency of the anterior

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mandibular region, which contained an impacted right mandibular canine with radiopaque foci. (Figure 2 and 3) The differential diagnosis was based on the clinical and radiographic findings included a dentigerous cyst, unicystic ameloblastoma, and Adenomatoid Odontogenic Tumor (AOT).

The surgical procedure consisted on firstly elevate a mucoperiosteal flap in order to expose the lesion (Figure 4). The bone window was performed to have access to the lesion (Figure 5). The lesion was separated from the surrounding bone and then, the lesion and impacted canine were enucleated (Figure 6, and 7). Intraoperatively, the specimen exhibited a solid and sand-like texture inside lesion (Figure 8).

The specimen was then sent to the department of pathology for histopathological study. The complete enucleation of the lesion was done under local anesthesia with the removal of impacted 43, and then the cystic cavity was completely cleaned (Figure 9).
Postoperative sutures were performed to finish the surgical intervention (Figure 10). 3 months Postoperative follow-up did not show any signs of recurrence or complications.

The histopathology of lesional tissue revealed a multilobular proliferation of spindle cells in sheets, duct-like pattern, and whorled arrangement of darkly staining epithelial cells suggestive of an odontogenic epithelial cells. The cuboidal to columnar cells arranged in the form of nests and rosettes. The duct-like structures with lumina of varying size were lined by columnar cells with palisading appearance. A few basophilic calcifications were also observed. The surrounding connective tissue stroma was less cellular in nature. Based on these findings, the final diagnosis was an adenomatoid odontogenic tumor (Figures 11).

**DISCUSSION**

AOT is a rare benign odontogenic neoplasm with predilection of occurrence in females and occurs mainly in the second decade of life as seen in our case. The lesions usually are characterized by the presence of an
asymptomatic jaw swelling, relatively small in size, and not exceeding 1-3 cm in diameter, as seen in our case.  

AOT exists in three clinical subtypes: follicular type (in 73% of AOT) which is intraosseous in location and is associated with an unerupted tooth (usually canine); the extra-follicular variant (24%) which is located intraosseously but is not associated with unerupted tooth, and the peripheral form which is the most rare (3%) and occurs within gingival mucosa.  

On radiographs, the intraosseous follicular variant of AOT shows a well delineated, unilocular radiolucency surrounding the crown of a retained tooth; a picture very similar to dentigerous cysts. Minor radiopacities around the retained tooth may be found in AOT and are considered a characteristic but not pathognomonic finding. About 2/3 AOT show distinct radiopaque calcification on radiographs.  

The extra-follicular variant presents as a unilocular, well-defined radiolucency seen between, above, or superimposed on the roots of erupted teeth and often resembles a cystic lesion.  

Our case presented as an extra-follicular AOT that was diagnosed clinically as a cystic lesion. AOT affects more commonly the maxilla than the mandible in a ratio of 2:1:1. The case reported here is considered uncommon as it involved the mandible.  

According to Philipsen et al., the fact that all AOT variants show identical histologic characteristics points to a common origin. The pathogenesis of this odontogenic tumor appears to involve persistence of remnants of the dental lamina, especially after odontogenesis of the successional lamina. These remnants give rise to epithelial rests that proliferate in response to an unknown stimulus.  

The radiographic findings of AOT frequently resemble other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumors, ameloblastomas, odontogenic keratocystic tumor, and periapical disease. Comparing diagnostic accuracy between intraoral periapical and panoramic radiographs, Dare et al. found that intraoral periapical radiographs allow perception of the radiopacities in AOT as discrete foci having a flocculent pattern within radiolucency even with minimally calcified deposits while panoramic often do not. Those calcified deposits are seen in approximately 78% of AOT.  

The diagnosis will especially be based on histological findings. At this stage also, this tumor must be differentiated from the calcified epithelial odontogenic tumor or Pindborg’s tumor which radiologically appears as a uni- or multilocular radiolucent image without peripheral dense border, scattered with radiopaque images of variable sizes and associated in 60% of the cases to an impacted tooth or an odontoma. Macroscopically, these tumors are perfectly limited by a fibrous connective capsule, having a smooth surface and a rather firm consistency, a tooth can be attached or included in the tumor. In the biopsy, a schematic liquid is frequently noted, and the significant growth gives an irregular shape to the cyst.  

Histologically, the adenomatoid odontogenic tumor is perfectly well-defined with the common presence of lobes and cystic cavities. Epithelial cells are closely aligned against the cyst border. There is little of the stroma. Cells have a vesicular nucleus and are arranged in nodules, spirals, and rosettes. They are crossed by canalicular structures lined with a cylindrical epithelium with rows of nucleus situated on the opposite side. Inside these canalicular cells, an eosinophilic amorphous material is found in contact with the apical pole of the cells. Calcifications are present in some places representing a sign of enamel formation. They are often in association with the groups of cylindrical cells morphologically identical to ameloblasts.  

Conservative surgical enucleation is the treatment modality of choice. For periodontal intrabody defects caused by AOT, guided tissue regeneration with membrane technique is recommended after complete removal of the tumor. Recurrence of AOT is exceptionally rare. Therefore, the prognosis is excellent. No recurrence was seen in our present case.  

CONCLUSION  

AOT is a relatively uncommon distinct odontogenic neoplasm, and is rightfully called as Perfect imitator of dentigerous cyst. It should be a part of differential diagnosis whenever we encounter a lesion in young patients with unerupted tooth (Two Third Tumor).  

REFERENCES  


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