

Case Report

Adenomatoid odontogenic tumor: An unusual presentation

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Abstract

Adenomatoid odontogenic tumor (AOT) is considered to be an uncommon odontogenic tumor which occurs most commonly in an unerupted maxillary canine. Some investigators consider it to be a benign neoplasm, while others consider it to be a hamartomatous malformation due to its limited size and to the lack of recurrence in most of the cases. The specific stimulus that triggers proliferation of progenitor cells of AOT is unknown, as in case of all odontogenic tumors. As it is exclusively associated with an unerupted or impacted tooth and the cytological resemblance to the dental lamina and enamel organ, it can be considered as odontogenic in origin. The present case highlights the unusual presentation of AOT which is not associated with an impacted or unerupted tooth.

Keywords: Benign neoplasm, odontogenic tumor, unerupted tooth

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INTRODUCTION

Adenomatoid odontogenic tumor (AOT) represents 3% to 7% of all odontogenic tumors, and more than 750 examples have been reported in the literature. Although this lesion was formerly considered to be a variant of the ameloblastoma and was designated as “adenoameloblastoma,” its clinical features and biological behavior indicate that it is a separate entity.^[1] In 1969, Philipsen and Birn proposed the widely accepted and the currently used name AOT.^[2] In 2005, the histological typing of the World Health Organization defined AOT as a tumor composed of odontogenic epithelium, which presents a variety of histoarchitectural patterns, embedded in mature connective tissue stroma, and characterized by slow and progressive growth.^[3]

AOT is a benign, painless, noninvasive, and slow-growing tumor that does not infiltrate the bone.^[4] Clinically, it is

often misdiagnosed as an odontogenic cyst. The tumor appears as an intraoral–extraoral swelling in the maxilla and is sometimes referred to as “two-third tumor” because it occurs in the maxilla in about two-third cases, about two-third cases arise in young females, two-third cases are associated with an unerupted tooth, and two-third affected teeth are canines.^[4,5]

The purpose of this article is to report an unusual case of AOT located in the maxilla which is not associated with an unerupted or impacted tooth, with an emphasis on radiographic findings and with pathologic correlation, and to review the existing literature on this tumor.

CASE REPORT

A 29-year-old male reported with swelling in relation to the right-side middle-third face for the past 2 months. The patient was asymptomatic 2 months back when he noticed a

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swelling in relation to the right-side middle-third face which progressively increased in size to attain the present size. The swelling was asymptomatic with no history of nasal stuffiness or nasal discharge. There was no history of trauma to that region. His medical history was not significant and he had no history of harmful habit. This was his first dental visit. A diffuse swelling of size approximately 3 cm × 3 cm was present in the right middle-third face extending superoinferiorly from the infraorbital rim to the corner of the mouth. Medially, the swelling extended up to the philtrum and laterally in line with the outer canthus of the eye. The skin over the swelling appeared normal with no evidence of secondary changes such as ulceration, sinus opening, or pus discharge [Figure 1]. On lymph node examination, a single left submandibular lymph node of size approximately 1 cm was palpable, which was soft-firm, mobile, and nontender.

On intraoral examination, a single swelling of size approximately 3 cm × 3 cm was present in relation to the right maxilla extending anteroposteriorly from the mesial aspect of 11 to the mesial aspect of 16. The palatal extension was 1 cm short of midpalatal raphae, and the swelling obliterated the buccal vestibule from 11 to 16. The mucosa over the swelling appeared normal with no evidence of secondary changes. On palpation, the inspector findings were confirmed. The swelling was firm to hard in consistency on the palatal aspect and soft to firm in consistency labially with tenderness on palpation [Figure 2].

Intraoral periapical radiograph in relation to 11, 12, and 13 revealed a well-defined homogeneous radiolucency extending from the periapical region of 11 to the mesial aspect of 14, and the complete extent was not covered [Figure 3]. Intraoral periapical radiograph in relation to 13, 14, and 15 revealed a well-defined corticated unilocular

homogeneous radiolucency extending from the mesial of 13 to distal of 15 with the displacement of 13 and root resorption in relation to 14 and 15 [Figure 4].

Maxillary cross-sectional occlusal radiograph revealed a well-defined corticated unilocular homogeneous radiolucency extending from the mesial of 11 to distal of 16 with buccal and palatal cortical expansion. Digital orthopantomogram revealed a well-defined corticated unilocular homogeneous radiolucency extending from the mesial of 11–17 with root resorption in relation to 14, 15, and 16. The floor of the right maxillary sinus could not be traced [Figure 5].

Computed tomogram axial bone window revealed a hypodense area involving the right maxilla with expansion of buccal cortical plate [Figure 6]. There was thinning of the buccal cortex with focal areas of destruction. Computed tomogram coronal bone window revealed a hypodense area involving the right maxilla with expansion of buccal and palatal cortical plate and lifting of the floor of the maxillary sinus. Computed tomogram three-dimensional reconstruction revealed cortical perforation and complete destruction in relation to the anterior wall of the maxillary sinus [Figure 7].

On the basis of history and clinical presentation, a provisional diagnosis of developmental odontogenic cyst most likely odontogenic keratocyst in relation to the right maxilla was given. The differentials included radicular cyst, dentigerous cyst, benign odontogenic tumors such as ameloblastoma and AOT, odontogenic myxoma, reactive lesions such as central giant-cell granuloma, central ossifying fibroma, and malignancies such as central mucoepidermoid carcinoma.



Figure 1: A diffuse swelling of size approximately 3 cm × 3 cm presents in the right middle-third face



Figure 2: Swelling of size approximately 3 cm × 3 cm presents in relation to the right maxilla with buccal and palatal expansion



Figure 3: Intraoral periapical radiograph in relation to 11, 12, and 13 revealed a well-defined homogeneous radiolucency extending from the periapical region of 11 to the mesial aspect of 14



Figure 4: Intraoral periapical radiograph in relation to 13, 14, and 15 reveals a well-defined corticated unilocular homogeneous radiolucency extending from the mesial of 13 to distal of 15 with the displacement of 13 and root resorption in relation to 14 and 15



Figure 5: Cropped digital orthopantomogram reveals a well-defined corticated unilocular homogeneous radiolucency extending from the mesial of 11–17 with root resorption in relation to 14, 15, and 16



Figure 6: Computed tomogram axial bone window reveals a hypodense area involving the right maxilla with expansion of buccal cortical plate

Thermal and electric vitality test revealed immediate response in relation to 21, delayed response in relation to 11 and 12, and no response in relation to 13, 14, 15, 16, and 17. On lesional aspiration, 5 ml of amber-colored fluid was obtained. Routine hemogram was within normal limits. Excisional biopsy was done, and the specimen was sent for histological investigation. Histologic investigation revealed a well-defined capsule and odontogenic epithelium which appeared in the form of strands and nodules and gives a whorled appearance, and there was calcification in some areas suggestive of AOT [Figure 8].

DISCUSSION

AOT is a slow-growing lesion, constituting about 3% of all odontogenic tumors followed by odontoma, periapical cemental dysplasia (cementoma), myxoma, and

ameloblastoma.^[6] They are largely limited to young patients, and two-thirds of all cases are diagnosed when the patients are 10–19 years of age. This tumor is definitely uncommon in a patient older than 30 years.^[11] The tumor is most often diagnosed in the second decade of life, and women are about twice as many affected than men. 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.^[7] These features are consistent with our present case except the gender variability where we have reported a young male. According to Philipsen and Reichart,^[8] AOT appears in three clinicotopographic variants: follicular, extrafollicular, and peripheral. The follicular and extrafollicular variants are both intrabony and account for approximately 96% of all AOTs of which 71% are of follicular type. The tumor is usually associated with unerupted teeth, frequently canines.^[9] However, in our case, there were no clinically missing teeth.



Figure 7: Computed tomogram three-dimensional reconstruction reveals cortical perforation and complete destruction in relation to the anterior wall of the maxillary sinus

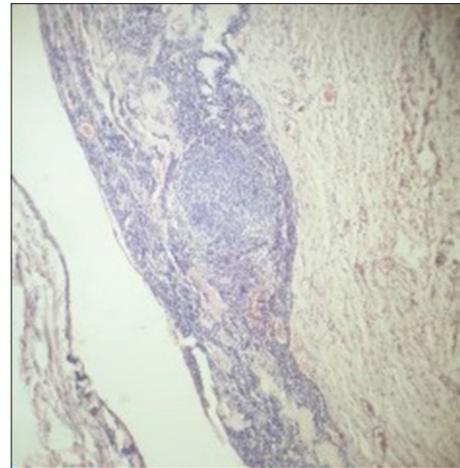


Figure 8: Histologic investigation reveals a well-defined capsule and odontogenic epithelium which appeared in the form of strands and nodules and gives a whorled appearance, and there was calcification in some areas

Radiographically, the periphery usually appears as a well-defined corticated or sclerotic border with internal radiopaque foci in two-thirds of the cases. One tumor may be completely radiolucent, another may contain faint radiopaque foci, and some may show dense clusters of ill-defined radiopacities although few multilocular cases have been reported.^[10,11] Radiographically, it should be differentiated from dentigerous cyst, which most frequently occurs as pericoronal radiolucency in the jaws.^[11] Root resorption is rare as the tumor enlarges and the adjacent teeth are displaced.^[9,10] In the present case, there was a well-defined homogeneous radiolucency without any calcific spots surrounded by a sclerotic border. Surprisingly, there was no evidence of any impacted teeth. Furthermore, there was severe resorption of the roots of premolars which is an unusual characteristic of AOT. AOT shows centrifugal expansion (uniform expansion in all directions). It has been hypothesized that at an early stage, AOT may expand the cortical plates, which within the cancellous bone spread linearly and then later may affect the cortical plates by expansion/resorption.^[12] Bicortical expansion was seen in our patient.

The histological findings for AOT reported in the literature reveal a remarkable similarity.^[7,13,14] The histological features of the tumor were described as a tumor of odontogenic epithelium with duct-like structures and with varying degree of inductive changes in the connective tissue. The tumor may be partly cystic, and in some cases, the solid lesion may be present only as masses in the wall of a large cyst.^[15] The tumor may contain pools of amyloid-like material and globular masses of calcified material.^[16] In our case, the histology was suggestive of well-defined capsule and odontogenic epithelium which appeared in the form of

strands and nodules and gives a whorled appearance. In some areas, calcification was noted.

Distinguishing from normal types of AOT, our present case had some unusual clinical, radiographical, and histopathological features. Those features include the following:

- AOTs are slow growing and relatively small in size but in our case showed unusual rapid growth to >3 cm within 1 month which is based on the history
- The radiographical features too revealed a unilocular radiolucency with distinct sclerotic borders without calcifications and associated with root resorption of premolars and first molar, which is unusual. To the best of our knowledge, only five cases of AOT with root resorption have been reported^[4]
- Furthermore, the unusual presentation of AOT which is not associated with any impacted or unerupted tooth has been reported. A similar case had been reported by Reddy Kundoor *et al.*^[17] in a 50-year-old male which explains the unusual aggressiveness of AOT.

Surgical excisional has been done, and the patient is under regular follow-up without showing any recurrence.

CONCLUSION

With respect to the presentation of the lesion which is not associated with any impacted or unerupted tooth and because of its aggressive nature, our case is a rare case of AOTs. AOT is commonly referred to as “two-third tumor,” but it does not mean that the tumor is restricted only to the two-thirds and some rare presentations do occur as in our present case. Thus, we conclude that AOT should

be considered as the differential diagnosis even for the aggressive lesions of the maxilla even if there is no evidence of clinically missing teeth.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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