

CASE REPORT

Adenomatoid Odontogenic Tumor as a Confusing Maxillary Gingival Swelling

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ABSTRACT

Maxillary gingival swelling encompasses a diverse group of lesions although clinically they are uncharacteristic. Adenomatoid odontogenic tumor (AOT) is one such uncommon cause of gingival swelling which is often misdiagnosed as other odontogenic lesions. AOT is a rare, slow growing, expansile, noninvasive, benign odontogenic epithelial tumor with relative frequency of 2.2 to 7.1%. It is predominantly found in young females, particularly in the second decade of life. It has a predilection for the anterior maxilla and frequently associated with unerupted permanent canines. AOT appears in three clinicotopographic variants: follicular, extrafollicular and peripheral. Histogenesis of AOT is uncertain. Some accept it as hamartoma while others argue in favor of true neoplasm. Treatment is conservative and the prognosis is excellent. We report a case of follicular variant of AOT in the right maxillary anterior region in a 14-year-old girl. The tumor was slow growing and was clearly encapsulated suggesting its benign behavior.

Keywords: Adenomatoid odontogenic tumor, Jaw swelling, Impacted canine.

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INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a rare tumor that comprises only 0.1% of tumors and cysts of jaw and 3% of all odontogenic tumors.¹ It is frequently encountered in young females, mostly in the second decade with a predilection for maxillary anterior region. Two-thirds of the cases are associated with unerupted canines.² It presents as symptom-free, slow growing, expansile lesion with a noninvasive character. The first description of AOT by Steensland dates back to 1905.³ The name AOT was coined by Philipsen and Birn in 1969 and it was adopted

by the first edition of the World Health Organization (WHO) classification of Odontogenic tumors in 1971.⁴ 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 degrees of inductive changes in the connective tissue.⁵ According to Philipsen and Reichart, there are three clinicotopographic variants of AOT: the follicular type (accounting for 73% of cases), which has a central lesion associated with an embedded tooth; the extrafollicular type (24%) has a central lesion and not associated with the tooth, and the peripheral variety (3%).⁶

In spite of the WHO description, there is no end to conflict regarding the histogenesis of AOT. Some describe it as benign while others have reported its aggressive nature.^{2,6,7} Here, we describe a case of Adenomatoid odontogenic tumor in a 14-year-old girl whose clinical, radiological, and histological features argue in favor of benign character.

CASE REPORT

A 14-year-old girl presented with a swelling over right upper jaw. She had noticed the swelling for last 2 months. The swelling was painless and growing slowly. There was no history of trauma. There was marked facial asymmetry. The right nasolabial fold was absent. The right ala and tip of the nose were deviated to left side (Fig. 1). It was a solitary swelling and there was no other swelling elsewhere in the body.

Intraoral examination revealed a 1.5 × 1 × 1 cm swelling on right maxillary alveolar region obliterating the labial vestibule and extending from right lateral incisor to premolar region (Fig. 2). The bony hard, nontender swelling had well-defined margins with hyperemic overlying mucosa. Teeth 12 and 14 were not responsive to electrometric pulp testing. There was retained 53 and 13 was missing.

Radiographic Features

Intraoral periapical radiograph (IOPAR) revealed a unilocular radiolucent image with a regular border and extending from 12 to 14 region and enclosing the crown of impacted 13. Occlusal radiograph showed a radiolucent area with a sclerotic outline. The root of the right lateral

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incisor was displaced mesially and crown distally, i.e. the usual orientation is disturbed (Fig. 3). The radiological picture is consistent with a cyst.

Other Investigations

Aspiration yielded 2.5 ml of straw colored fluid. Routine hematological investigations were done and were within normal limits.

Macroscopic picture: Under the clinical diagnosis of dentigerous cyst.

The tumor was enmass enucleated and 53, 12, 14 along with impacted 13 extracted (Figs 4 and 5). The tumor was bluish red in color measuring approximately 1.5 × 1 × 1 cm. The labial cortex was very thin and had several areas of complete resorption. The specimen was sent for histopathological examination. On grossing, it revealed a thin-walled cyst with whitish necrotic material.

Microscopic picture: (Figs 6 to 8).

- Polyhedral to spindloid cells with scanty cytoplasm scattered in fibrocollagenous stroma. The cells were arranged in ductal pattern, nests as well as whorls.

- Occasional small tubules lined by columnar cells arranged in rosette pattern.
- Eosinophilic coagulum among the cells.
- The lesion was encapsulated with areas of hemorrhage around the capsule.

The pathologist’s impression was adenomatoid odontogenic tumor.

Follow-up: Radiograph (at 6 months) shows well-circumscribed radiolucency with radiopaque margin measuring almost 1 × 1 cm (Fig. 9) and no recurrence till date (Fig. 10).

DISCUSSION

The site of occurrence of AOT is greater in maxilla (65%) than in mandible (35%). In 74% of the cases, the tumor is associated with an unerupted tooth and in over two-third of cases, it is the maxillary or mandibular cuspid.² The expansile nature of the tumor often causes displacement of the neighboring tooth as was seen with the lateral incisor in the present case.



Fig. 1: Frontolateral view of the patient showing prominently the facial asymmetry on the right side of the face with obliteration of the nasolabial fold, right ala and tip of the nose deviated to the left side



Fig. 2: Intraoral photograph showing a bulged-out lesion in the maxillary anterior right alveolar region obliterating the labial vestibule

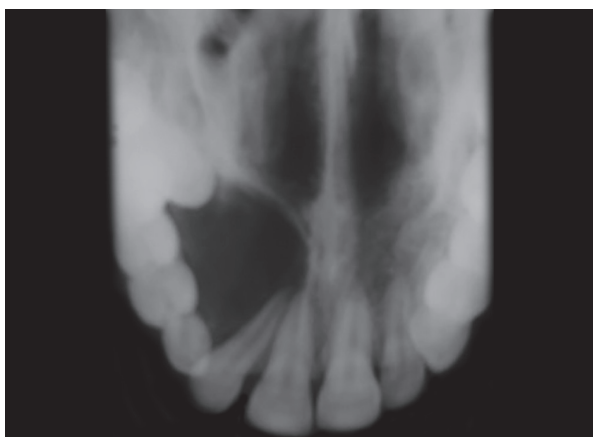


Fig. 3: Maxillary cross-sectional occlusal radiograph showing a well-defined radiolucency in the anterior segment with impacted right maxillary canine with divergent root of lateral incisor



Fig. 4: Peroperative photograph showing ballooning of the lesion in the bony cavity

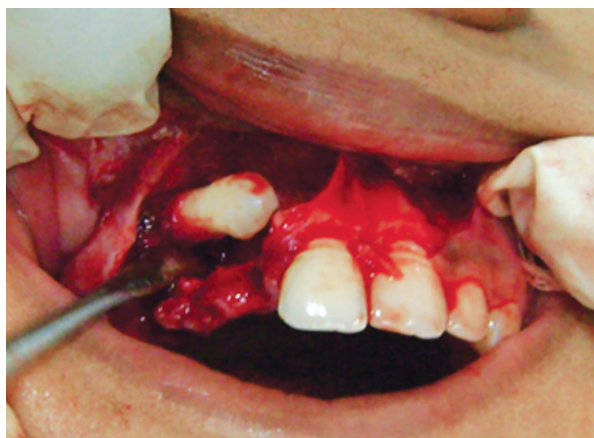


Fig. 5: Surgical exposure of the impacted permanent maxillary canine

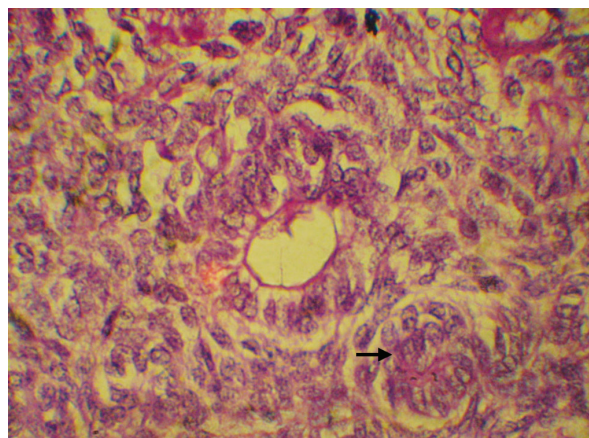


Fig. 6: Photomicrograph showing the columnar cells arranged in ductal pattern with a central lumen cells arranged in rosette pattern (→) (H&E stain: 40x)

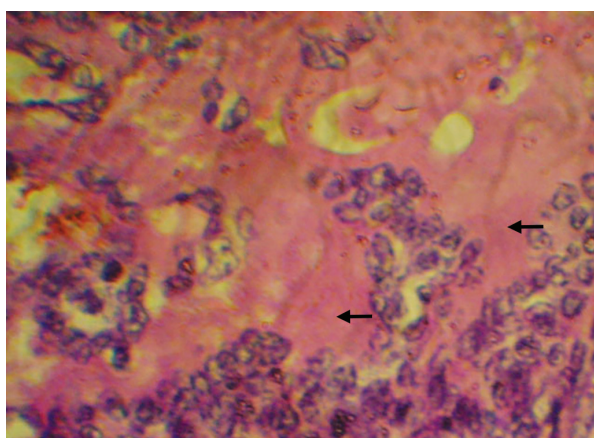


Fig. 7: Islands containing pools of amorphous eosinophilic coagulum (→) (H&E stain: 40x)

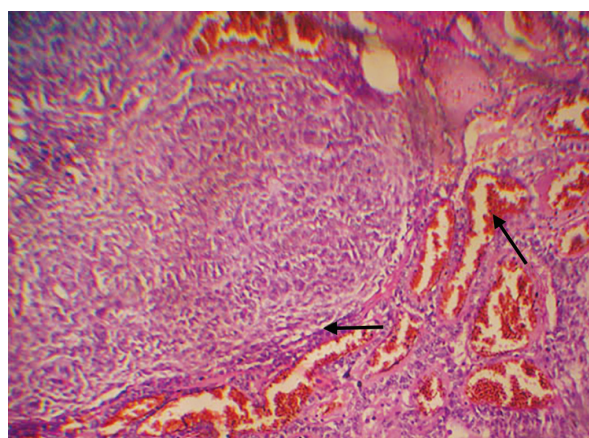


Fig. 8: Photomicrograph showing encapsulated lesion and areas of hemorrhage around the capsule (→) (H&E stain: 10x)

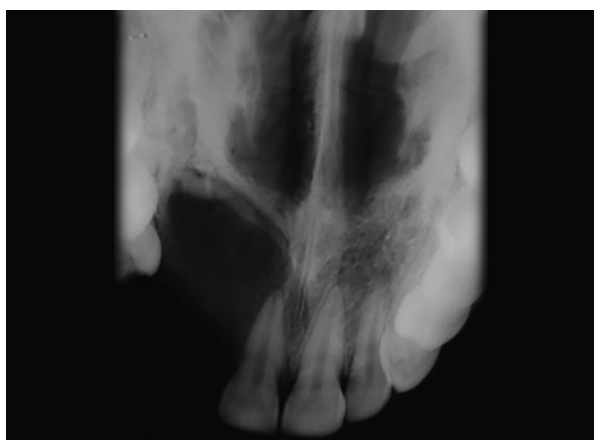


Fig. 9: Postoperative occlusal radiograph showing bony cavity with absence of impacted canine



Fig. 10: Postoperative frontal view showing disappearance of the facial deformity

Most AOT occur intraosseously as either the follicular or extrafollicular type. The third variant, the peripheral type, is a rare form that arises in the gingival tissue and is often preoperatively classified as a gingival fibroma or epulis attached to the labial gingiva.

There has been description of as many as 20 different histological patterns of AOT.⁸ AOT frequently resembles

dentigerous cysts. Radiologically, an AOT often appears to envelop the crown as well as part of the root of an unerupted tooth, whereas dentigerous cysts do not envelop the roots. Other odontogenic lesions, such as calcifying epithelial odontogenic tumor, odontogenic keratocysts, calcifying odontogenic cysts, ameloblastoma, ameloblastic fibroma, ameloblastic fibro-odontoma are

differentiated from AOT on the basis of their histological characteristics.

Most authors accept the odontogenic origin of AOT. It occurs within the tooth bearing areas of the jaws, found in close association with embedded teeth, having cytological features similar to those of the various components of the enamel organ, dental lamina, reduced enamel epithelium and/or their remnants.⁹

Formation of duct-like structures is due to epithelial component of the lesion. Foci of calcification are often seen scattered throughout the tumor and these have been interpreted as attempted enamel formation. Ultrastructural study also reveals similarity of the lesion to the normal enamel organ.

Whether AOT is a hamartoma or neoplasm remains controversial. Since, it resembles odontogenic source, is of limited size in most cases and there is lack of recurrence many authors believe it to be a hamartoma.^{2,6} There are some reports in which the tumor attain large size.⁹ WHO Classification (1971) stated 'It is generally believed that the lesion is not a neoplasm.' But, there are reports where the tumor had rapid growth and lacked well defined fibrous capsule suggesting aggressive behavior.^{9,10}

However, in the present case, the tumor was slow growing with a well-defined fibrous capsule and no increase in mitotic activity. There was healing of the lesion following resection and no recurrence till date

(followed up for 3 years). All these features are suggestive of the benign nature of this lesion.

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