Case Report

Adenomatoid Odontogenic Tumor of Maxilla: A Case Report

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is a distinct odontogenic tumor that is entirely odontogenic epithelium in origin which accounts for about 3-7% of all odontogenic tumors. It is a benign (hamartomatous), noninvasive lesion with slow but progressive growth. It is primarily found in young and female patients, located more frequently in the maxilla in most cases associated with an unerupted permanent tooth. Treatment is conservative surgical excision, and the prognosis is excellent. Here, we report a case of AOT of the maxilla in a young girl aged 16 years. A case of AOT of the maxilla is reported with unusual features such as large size and aggressive behavior. The unique radiological manifestations of the lesion helped in the diagnosis, and it was managed conservatively with no evidence of recurrence.

Keywords: Adenomatoid odontogenic tumor, Aggressive behavior, Benign, Odontogenic epithelium, Odontogenic tumor

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INTRODUCTION

Adenomatoid odontogenic tumor (AOT), an uncommon benign epithelial lesion of odontogenic origin, was first described in 1907 by Dreibladt, as a pseudoadenoameloblastoma.¹² AOT was first reported by Harbitz in 1915 as cystic adamantoma.³ Philipsen and Birn proposed the widely accepted and currently used name AOT, a term that was adopted by the first edition of the World Health Organization (WHO) classification of odontogenic tumors in 1971.⁴

AOT is also called “two-third tumor,” because two-third occur in young females as well as in maxilla, two-third of the cases are associated with unerupted teeth, and two-third of the affected teeth are canines.⁵⁶ 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 AOT is subdivided into three groups by Philipsen et al. and referred to as follicular, extrafollicular, and peripheral.⁴⁵ All the variants have common histological characteristics as well as indicative of a common origin as a complex system of the dental lamina or its remnants. The follicular and extra follicular variants comprise 96% of all AOT, and 71% of these are the follicular variant.⁷ The follicular variant is associated with the crown and frequently part of the root of an impacted or unerupted tooth. The majority of the cases, comprise of about 88%, are diagnosed in the second and third decades of life. The tumor may be partially cystic, and in a few cases, the solid lesion may be present as masses in the wall of a large cyst. The epithelial lining of the odontogenic cyst may transform into an odontogenic neoplasm - like an ameloblastoma or AOT. While most of AOT arises in anterior maxilla, it can rarely also originate in the wall of a dentigerous cyst of the maxillary antrum and very rarely in posterior maxilla with an impacted second molar.⁴⁶ Conservative surgical enucleation is the most suggested choice of treatment.⁷ Recurrence rate for AOT is exceptionally rare.⁷ Here, we are presenting a case of AOT of the maxilla.

CASE REPORT

The 16-year-old female child reported to the Department of Oral Pathology with a complaint of swelling on left side of upper jaw since 1 year. History of the present illness 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. Extra oral examination revealed mild facial asymmetry with the obliteration of the nasolabial fold. The swelling was extending from ala of the nose to malar prominence. Intraoral examination revealed a solitary diffuse swelling was present at buccal vestibule extending from 11 to 17; 6 cm × 4 cm × 4 cm in diameter. Obliteration of buccal vestibule with the expansion of buccal cortical plate is seen. Overlying mucosa was whitish, shiny in color. On palpations, all inspectory findings are confirmed. The swelling was hard in consistency, firm, non-tender having well-defined borders, smooth surfaces, and fluctuations were noted (Figure 1).

Orthopantomogram of the patient showed well-defined unicystic radiolucency seen in relation to 23, 24 (Impacted teeth) measuring about 2 cm × 1 cm in size extending from 21 to 26. The floor of the maxillary sinus is pushed by the lesion in a superior direction. Displacement of teeth (21, 22, 25, 26) (Figure 2).

Lesional tissue was removed completely by surgical excision, and the specimen was sent for the histopathological examination (Figure 3). 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 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, a histopathological diagnosis of AOT was made (Figures 4 and 5).

DISCUSSION

The AOT is an unusual odontogenic tumor which causes jaw swelling. There is a slight female over male predilection which is almost 2:1 and it appears most often in the second decade of life. In general, the tumors do not exceed 1-3 cm in maximum diameter, but
they can be larger. Three clinicopathologic variants of the tumor have been described: - follicular type (73%), associated with an impacted or embedded tooth; extra follicular type (24%), no connection with the tooth; and the peripheral variety (3%). All these variants have identical histological features. In our case; there were impacted canine and premolar associated with the lesion, so the diagnosis of follicular was given.

Radio graphically most AOTs have a well demarcated unilocular radiolucency which usually exhibits a smooth corticated and occasionally sclerotic border. Most lesions are pericoronal or juxtacoronal, but the radiolucency might extend apically beyond to cemento-enamel junction on at least one side of root. About 65% of the cases demonstrate faintly detectable radiopaque foci within the radiolucent lesion. Divergence of roots and displacement of teeth occurs more often than root resorption.

The dispute of whether AOT is an anomalous developmental hamartomatous growth or a true benign tumor has not been settled yet. Hypothesis of AOT as hamartoma is due to the limited size of most cases and the lack of recurrence. While as a tumor is because of the belief that the limited size of most cases origin from the fact that most are detected early after a routine radiograph and detached before the slow growing tumor reaches a clinically noticeable size.

The origin of AOT is also controversial. Some believe they originate from the odontogenic epithelium of a dentigerous cyst. In addition to the anterior maxilla, the tumor has been reported in posterior mandibular areas, for example, the angle of the mandible. Therefore, dental lamina remnants probable represent the progenitor cells for this benign odontogenic tumor. According to this supposition, the lesion grows next to or into a nearby dental follicle, leading to the “envelop mental theory.”

In our case, the lesion surrounded a fully formed canine tooth, which suggests “envelop mental” pathogenesis. Recent reports indicate that the cells of an AOT usually differentiate toward an apparent ameloblastic phenotype but fail to achieve further functional maturation.

The most frequent histological pattern is a proliferation of nests, sheets, and cords of ameloblast-like cells supported by a scanty often hemorrhagic stroma. These cells may be structured to form whorls, rosettes, nodules or surround ovoid spaces to form duct-like structures. Moreover, “amyloid” like amorphous eosinophilic material may be lining the ductal lumens or likely as intercellular droplets in cellular areas. Calcific material is often found throughout the tumor and appears to develop primarily as the junction between tumor epithelium and adjacent vascular stromal tissue.

Conservative surgical enucleation is the most suggested choice of treatment. The recurrence rate for AOT is exceptionally rare. The prognosis is excellent when completely removed in toto.

CONCLUSION

The tumor is well-encapsulated and showed an identical benign behavior. Therefore, conservative surgical enucleation produces excellent outcome without recurrence. Our patient has been under follow-up for 6 months and was rehabilitated with fixed prosthesis.

REFERENCES


Source of Support: Nil, Conflict of Interest: None declared.