ADENOMATOID ODONTOGENIC TUMOUR- A CASE REPORT

Vivek Kumar Rai, Vijay Agrawal, Manju A Nair, Gaurav Das

ABSTRACT
The adenomatoid odontogenic tumour (AOT) is a rare odontogenic tumour often misdiagnosed as an odontogenic cyst. Treatment of choice for AOT is conservative surgical excision or enucleation. Recurrence is rare with good prognosis. This paper reports the surgical management of AOT in right maxillary anterior of a 20-year-old man. 

Keywords: Adenomatoid odontogenic tumour; odontogenic tumour; Maxilla

Introduction
The adenomatoid odontogenic tumour (AOT) is a rare odontogenic tumour constituting only 3-7% of all the odontogenic tumors and first described by Drieibaldt in 1907. Based on Histological typing of odontogenic tumors by World Health Organization (WHO), adenomatoid odontogenic tumor is defined as “A tumor of epithelium with duct-like structures and with varying degrees of inductive change 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”.  

Topographically, the AOT occurs as peripheral and central variants. There are three variants of adenomatoid odontogenic tumor, follicular variant (73%) has a central lesion associated with an embedded tooth, extrafollicular variant (24%) has a central lesion and peripheral variety (3%) has no connection with the tooth. Pathogenesis of AOT is controversial, the lesion originates from odontogenic epithelium (enamel organ or dental lamina remnants) with inductive influence on odontogenic ectomesenchyme and consequent production of dentinoid material. Some believe they originate from the odontogenic epithelium of a dentigerous cyst. Radiographically, the tumour usually appears as a well-circumscribed, unilocular radiolucency, which may be associated with an unerupted tooth, most often a canine. This paper reports the surgical management of AOT in right maxillary anterior region of a 20-year-old man.

Case Report
A 20 years old male patient reported to the Department of Oral and Maxillofacial Surgery with the complaint of painful swelling present in the right maxillary canine region for last one month. History revealed that, the swelling was gradually increasing in size causing discomfort in the affected region. His past medical and dental history was not significant. On clinical intraoral examination a firm, rounded and tender swelling, present in the right maxillary region. The swelling was extending anteriorly from the distal surface of maxillary right central incisor to the distal surface of maxillary right second molar tooth posteriorly (Figure 1) with a palatal swelling. On palpation the swelling was firm, tender, smooth surfaced, non fluctuant and non compressible. Right central incisor was supra-erupted. Tooth mobility was noticed. Orthopantomogram revealed large, round shaped, well defined, radiolucent area with impacted permanent right maxillary canine, causing bone resorption, present between the of right maxillary central incisor and second molar tooth with evidence of root resorption of deciduous canine and first permanent molar. OPG revealed a mesioangular impaction of right maxillary third molar tooth, which was non-symptomatic on clinical examination (Figure 2).

CT scan revealed a lesion of 3.7cm x 4.0cm x 4.2cm in the right maxillary bone and causing expansion of bone. Superiorly the lesion is seen to bowing the anterior and medial wall of the maxillary sinus. Medially the lesion causes compression on the lateral wall of nose resulting in narrowing of nasal cavity. Inferiorly it causes thinning and expansion of superior alveolar arch extending up to the midline and involving the hard palate (Figure 3).

Based on history and clinical presentation, a provisional diagnosis of AOT was made. The differential diagnosis includes dentigerous cyst. The complete hemogram of the patient was within normal limits. Incisional biopsy was performed under local anesthesia, and the biopsy specimen constitutes of both soft and hard tissues. Histopathological examination shows a thick fibrous capsule enclosing spindle shaped cells arranged in whorl-like pattern and numerous duct like structures lined by cuboidal cells enclosing eosinophilic amyloid like material with Basophilic calcifications, suggestive of adenomatoid odontogenic tumour (Figure 2).

Surgical enucleation under general anesthesia was planned and consent was obtained. Followed by the pre anesthetic evaluation and preoperative preparations, patient was shifted to the operation theatre for surgical enucleation under general anesthesia. General anesthesia was established by nasoendotracheal intubation through left nostril. Painting and draping performed following the standard protocol. Crevicular incision placed from 21 to 16 and complete mucoperiosteal flap reflected very carefully. Complete enucleation of lesion was performed (figure V). The enucleated surgical specimen was measured 4.0cm x 4.0cm x 3.5cm (figure VI). Wound closure was done by simple interrupted suture using 3-0 silk. The postoperative healing was uneventful.
Discussion

Adenomatoid odontogenic tumor is a slow growing lesion, constituting only 3% of all odontogenic tumors with a pre-dilection for the anterior maxilla. Rick et al have reported adenomatoid odontogenic tumor to occur with many types of cysts and neoplasm's including dentigerous cyst, calcifying odontogenic cyst, odontoma and ameloblastoma. 69% of adenomatoid odontogenic tumors are diagnosed in the second decade of life, and more than half occur during the teenage years. Generally the tumors do not exceed 1–3 cm in greatest diameter, but they can be larger. The lesions are typically asymptomatic, but growth of the types with central lesion results in cortical expansion. The involved teeth are commonly impacted, and adjacent teeth may be slightly displaced. Displacement of neighboring teeth due to tumor expansion is much more common than root resorptions. Many different names like adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum or teratomatous odontoma have been used in the past to define the lesion, but currently it is called as, AOT.

The origin of AOT is controversial. However, evidence also exists that the tumor could be derived from epithelial remnants of the dental lamina complex system. On radiographs; the intra osseous follicular variant of AOT shows a well-delineated, unilocular radiolucency surrounding the crown of a retained tooth, a picture indistinguishable from follicular cysts. Indeed, the radiological findings of AOT frequently share characteristics of other odontogenic lesions such as dentigerous cyst, calcifying odontogenic cyst or tumor, ameloblastoma, keratocystic odontogenic tumor, or periapical disease.

At low magnification the most striking pattern is that of various sizes of solid nodules of follicular or cuboidal epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue. Between the epithelial cells of the nodules and in the centre of the rosette-like configuration is found eosinophilic amorphous material, often described as tumour deposits. Conscious within the cel-ular areas are structures of tubular or duct-like appearance.

All variants of AOT are well encapsulated and show an identical benign behavior. Conservative surgical enucleation or curettage is the treatment of choice with only rare recurrence. For periodontal intrabony defects caused by AOT, guided tissue regeneration with membrane technique is suggested after complete removal of the tumor. The patient we described in this case report is healthy without recurrence and is under follow-up after local excision. Our case report supports the general description of adenomatoid odontogenic tumor in the previous studies. There is an important need to report similar and other such cases, as we feel that many cases are surgically managed but unfortunately not reported. All such cases should be reported by the maxillofacial clinicians, so that we can increase not only our local literature bank, but also play a positive contribution to our expanding and demanding maxillofacial specialty.

Conclusion

In conclusion, the rarity of adenomatoid odontogenic tumor may be associated with its slowly growing pattern and symptomless behavior. Therefore, it should be distinguished from more common lesions of odontogenic origin in routine dental examinations.

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How cite this article

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Source of Support: Nil
Conflict of Interest: None Declared