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Case Report

Maxillary Adenomatoid Odontogenic Tumor - An Enigmatic Case Report

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Abstract

Adenomatoid odontogenic tumor is one of the uncommon tumor of oral cavity which accounts for nearly 3-7% of all the odontogenic tumors. Here we report an enigmatic case of AOT in maxilla in a young female of 24years describing its surgical management, clinical and morphological characteristics. Initially case was provisionally diagnosed to be as globulomaxillary cyst based upon its clinical and radiological findings.

Keywords: Adenomatoid Odontogenic Tumor; Maxilla; Tumor

Introduction

Adenomatoid odontogenic tumor (AOT) is a rare benign oral lesion affecting younger individuals with an impacted tooth, mostly canine. AOT constitutes for 3.1% of all the odontogenic tumors [1,2].

Regarding its etiopathogenesis, the lesion originate from odontogenic epithelium (enamel organ and dental lamina remnants) [3].

Radiographically it appears to be a well circumscribed unilocular radiolucent area found associated with impacted tooth most frequently associated tooth is canine [4]. Topographically, AOT have peripheral and central variants, the latter is further divided into follicular type(embedded tooth) and non- follicular type (without embedded tooth). The percentage for central variant accounts for 97.2% out of which 73% are follicular type. A study showed that follicular AOT was found to be associated with the embedded tooth is 93.2%. Among all four of the permanent canines, the maxillary

canine accounts for 41.7% of AOT associated with embedded teeth [5].

The histogenesis of AOT is not certain yet it is sometimes characterised as hemartomatous lesion of the oral cavity. AOT is often reffered to be as "Two Thirds Tumor" as it occurs in 2/3rd cases involving maxilla, affecting 2/3rd cases in young females associated with impacted tooth in 2/3rd cases with 2/3rd of canines involved [6].

Through this case report we review the literatures describing adenomatoid odontogenic tumor its clinical, radiological and histopathological features with enigma in identifying this tumor.

Case Report

A 24year old female patient reported to the clinic presenting with the chief complaint of painful swelling in right upper front tooth region since 1-2 months. Her past dental and medical history were found to be insignificant. Upon intraoral examination a firm, smooth, round and tender swelling was found. Size ranging of about 2-3 cm, with well defined margins obliterating the buccal vestibule.

The teeth associated with the lesion were found to be intact, however lateral incisor and canine were found tilted.

Patient's CBCT showed a pear shaped well defined radiolucency with bone resorption found between the diverging roots of involved teeth i.e. maxillary lateral incisor and canine (Figure 1.a,b).

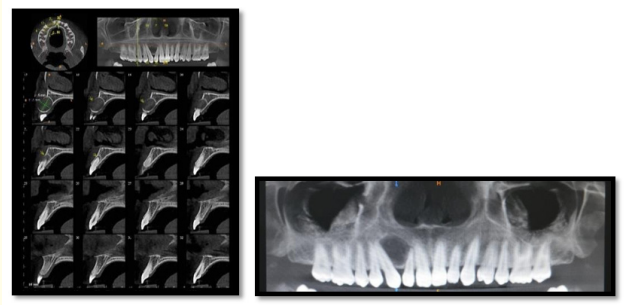


Figure 1: a,b. CBCT showing pear shaped radiolucency in between maxillary right lateral incisor and canine roots).

As based upon the clinical as well as radiological findings, provisional diagnosis of glubulomaxillary cyst was made.

Treatment: Routine pre operative procedure was done with the informed consent to the patient about the planned surgical procedure, enucleation of suspected cystic pathology was completed.

A full thickness mucoperiosteal flap was raised extending from the right maxillary lateral incisor up to right maxillary 1st premolar.

The small bony window was enlarged and the lesion was successfully enucleated as in whole to prevent any remnants (Figure 2).

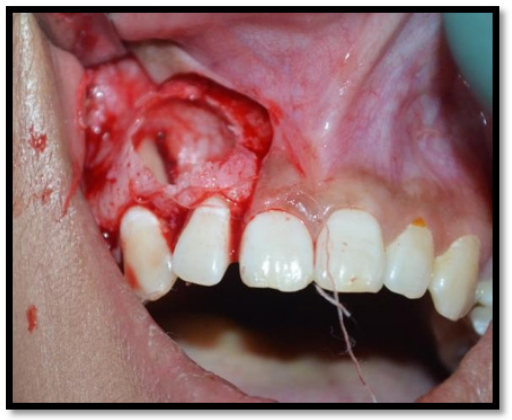


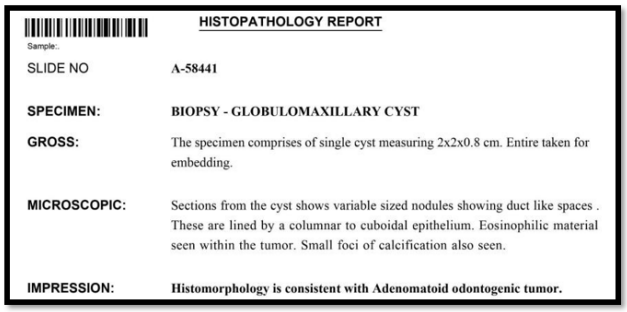
Figure 2: Full thickness mucoperiosteal flap raised with enlargement of bony window.

The excised specimen was then sent for histopathological examination which in result showed variable sized nodules with duct like spaces lined by columnar and cuboidal epithelium containing eosinophilic material. Histomorphology remained consistent with adenomatoid odontogenic tumor (Figure 3).

The healing remained uneventful and fortunately follow up visits were not lined with any kind of post operative complications.

Discussion

The case of AOT first seemed to fit to the clinical and radiological features of globulomaxillary cyst, which finally was diagnosed



HISTOPATHOLOGY REPORT

Sample:

SLIDE NO	A-58441
SPECIMEN:	BIOPSY - GLOBULOMAXILLARY CYST
GROSS:	The specimen comprises of single cyst measuring 2x2x0.8 cm. Entire taken for embedding.
MICROSCOPIC:	Sections from the cyst shows variable sized nodules showing duct like spaces . These are lined by a columnar to cuboidal epithelium. Eosinophilic material seen within the tumor. Small foci of calcification also seen.
IMPRESSION:	Histomorphology is consistent with Adenomatoid odontogenic tumor.

Figure 3: HPE report showing final impression of AOT.

to be adenomatoid odontogenic tumor on the basis of HPE report after total surgical removal of the lesion as a chair side procedure which was done under local anesthesia.

AOT is one of the uncommon as well as distinct odontogenic neoplasm which was first described by Steens land in 1905 [7].

However variety of terms has been coined describing this rare tumor. One of the author Unal et al produced a list with all the nomenclatures which are being reported for AOT in the literatures so far. Various names like admantinoma, adenoameloblastoma, adenoblastic adenomatoid tumor, epithelioma adamantinum of teratomatous odontoma were been used terms in past for defining this lesion, but now a days it is called as AOT i.e. adenomatoid odontogenic tumor [8]. Age distribution shows its occurrence in 2/3rd cases in 2nd decade of life and more than half in teens, predominance in females is there. Female: male ratio is 1.9:1 [9]. In this case too female patient was reported of age 24 years.

The AOT is predominantly found affecting the maxilla. The maxilla: mandibular ratio ranges from 2.6:1 [10]. In this reported case lesion was found affecting the maxilla only.

The clinical features generally have chief complaint of missing tooth, it usually presents with asymptomatic swelling which is slow growing associated with impacted tooth. However, its rare peripheral variant often occurs in gingival tissue of tooth bearing area [10].

AOT has similar radiographic features resembling few others odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumors, globulomaxillary cyst, ameloblastoma, odontogenic keratocysts and peripheral lesions [11].

The follicular variant show well circumscribed unilocular radiolucent area found associated with the crown and root of an unerupted tooth whereas the extra follicular is located above or between or may be superimposed over the roots of erupted tooth [12].

Displacement of adjacent teeth are often found associated with lesion due to expansion of tumor as compared to their root resorptions [5]. Our case also has root of adjacent teeth displaced due to growth of the lesion.

WHO defined AOT with duct like structures with presence of varying degree of changes in connective tissue tumor, sometimes it may be partially cystic while in some of the cases solid lesion could be found as a large cyst [13]. Eosinophillic, uncalcified, amorphous material can be found in its microscopic findings which are called as “tumor droplets” [14]. Our case showed duct like structures in histopathologic findings with presence of eosinophilic material.

Conservative form of surgical enucleation is the treatment of choice, AOT have rare chances of reoccurrence [15]. which makes its prognosis to be excellent when removed completely. Maxillofacial surgeons must report such cases, for not just increasing literature bank but also contribution and awareness be created in diagnosing such rare entity.

Conclusion

AOT is one of the uncommon odontogenic tumor, found primarily in young females. This case describes a rare entity of adenomatoid odontogenic tumor which is an extra follicular type variant. It was provisionally diagnosed as globulomaxillary cyst based upon clinical and radiographic findings but was finally diagnosed to be as AOT based upon histopathologic findings. There is an important need to report such cases as many of similar cases are being surgically managed but unfortunately not reported and go unnoticed.

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