



A RARE HYBRID AOT OF MAXILLARY ANTRUM: A CASE REPORT WITH 10 YEARS FOLLOW-UP

Oral Pathology

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ABSTRACT

Introduction: Amongst all odontogenic tumors adenomatoid odontogenic tumor (AOT) accounts for only 3% of them. Being odontogenic in origin, the tumor shows a plethora of histoarchitectural patterns.¹ Sex predilection is towards females and likely occurs in second to third decades of life. Anterior maxilla is the commonest region for AOT; maxillary antrum being rare; in conjugation with dentigerous cyst (DC) then being the rarest.² **Observation:** Here we describe a case of gigantic AOT in 17-year-old male that occupied the left maxillary antrum with an impacted tooth which was clinically diagnosed as dentigerous cyst. In toto removal of the cystic tumor was accomplished with removal of retained impacted canine as well. Histopathological examination inferred the presence of an AOT with dentigerous like cystic lining. With uneventful healing no local recurrence was observed after 10 years of follow up. **Conclusion:** Implied take away lesson the prime importance of histopathological evaluation in all enucleated cysts, simultaneous more research into fourth type of a "hybrid variant" of AOT.

KEYWORDS

Adenomatoid Odontogenic Tumor, Dentigerous cyst, Maxillary sinus, case reports

INTRODUCTION

Adenameloblastoma, being the old name for adenomatoid odontogenic tumor (AOT) was priority mentioned in 1907 by Dreidblad. Further in 1948 Stafne elaborated its description according to his experience. Almost after 20 years, in 1971 World Health Organization (WHO) acquired "adenomatoid odontogenic tumor" for explaining this kind of entity. The idea of providing such term was of Philipsen and Birn.^{1,2} The latest edition of WHO classification in 2005 included, AOT in odontogenic tumors classification under the section of tumors having odontogenic epithelium without ectomesenchyme. Predilection of female: male ratio is 1:1.9 with region predilection for anterior maxilla. The tumor is clinically seen as a slow progressive growing asymptomatic swelling usually involving an unerupted tooth.^{3,4}

Other than AOT another developmental odontogenic cyst that encapsulates the crown of unerupted teeth is a Dentigerous cyst (DC). These cysts are formed by expansion of follicles of unerupted teeth, and are always connected to the cemento-enamel junction of tooth. Widely accepted theory for development of cysts is by accumulation of fluid between the reduced enamel epithelium and the tooth crown. They can also occur from remnants of odontogenic epithelium as well.⁵ The transformation of DC into AOT is extremely rare. According to literature the epithelial lining of the odontogenic cyst may convert into an odontogenic neoplasm-like ameloblastoma or AOT. Such pathologies are rare of all; and associating maxillary antrum are few countable on fingers.⁶

The aim of this case report is to make an account of an infrequent entity seen in a 17-year-old male patient, intruding the left maxillary antrum with an impacted canine. This report also adds that the clinical nature of the lesion displayed features of dentigerous cyst but histopathologically came out to be an AOT. Further the authors have also discussed clinical, radiographic, histopathologic and therapeutic features of the case.

CASE REPORT:

A 17-year-old male patient reported to oral surgery department with a chief complaint of pain and slow growing swelling causing facial asymmetry on the left side of the face. History revealed that the swelling started one year ago and gradually increased in size. Patient also gave history of habit of gutkha chewing since 5 years in frequency of 10 pouches per day. However, the patient had neither consulted nor sought treatment for the same prior to this presentation. There was no

previous history of fever, tenderness over the sinus area, trauma or injury to the maxillo-facial region.



Figure 1: Extra-oral and intra-oral examination of the lesion

Figure 1 depicts the intraoral and extra oral status of the patient. On intra-oral examination, the left upper permanent canine was missing with a retained deciduous left canine intact. Buccal vestibular obliteration was visualized extending from the left upper lateral incisor to the second premolar of the same arch. Clinically the swelling measured approximately about 3 x 3 cm in diameter with a positive fluctuation on the buccal surface. The swelling seemed to have caused expansion of both the buccal and palatal cortices. Extraorally, there was apparent facial asymmetry of the left upper side of the face as seen in the figure.

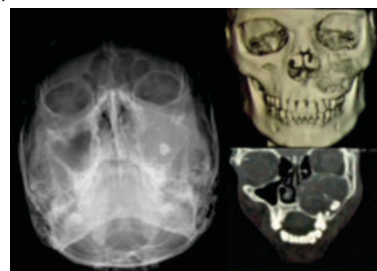


Figure 2: Radiographic investigations of the lesion

An occipito-mental view (Water's view) of para-nasal sinus radiograph revealed a well-defined radiolucency within the left maxillary sinus region extending from the canine to premolar region. The 3dimensional computed tomography revealed considerable thinning of the buccal cortical plate. All these radiographic investigations are shown in **Figure 2**. The clinical and radiographic presentations resulted in the provisional diagnosis of dentigerous cyst. Further fine needle aspiration cytology was undertaken as a

diagnostic tool. The fluid was straw colored with the protein estimation of 7.79gm/dl suggestive of Dentigerous cyst. Hence in total enucleation of the lesion was planned. Following routine blood works, the cystic lesion was enucleated under general anesthesia, by placing a vestibular incision from left upper lateral incisor to the second premolar region. Full thickness muco-periosteal flap was raised to expose the lesion on the anterior wall of the maxillary sinus. A cyst-like mass was enucleated along with its lining and contents, which included an impacted canine as well. **Figure 3** explains the vestibular incision, the fluid retrieved and in toto enucleated cystic mass from the disease.



Figure 3: In Total Excision Of The Pathology With Fluid Content From The Lesion

The cavity was irrigated, hemostasis was achieved, and sutures were placed to approximate the incision. The enucleated cystic lining and contents were subsequently submitted for histopathological examination. Sutures placed were removed after 1 week post operatively with uneventful healing. On gross examination the lesion revealed a cystic lining with black to brown roughened borders with an impacted tooth in the cavity filled with greenish colored cheesy material. Histopathology report revealed that cystic lining was composed of two - three layers of non-keratinized squamous epithelial cells with fibrous connective tissue and areas of chronic inflammatory cell infiltrate underlying it suggestive of dentigerous cyst. The tumor proper showed spindle shaped cells surrounding the duct like structures forming a rosette pattern suggestive of AOT. Hence the final diagnosis came out to be an AOT with dentigerous cystic lining involving an impacted canine. This can be seen in **Figure 4** which shows up the gross and histopathological examination views of the mass involving an impacted canine.

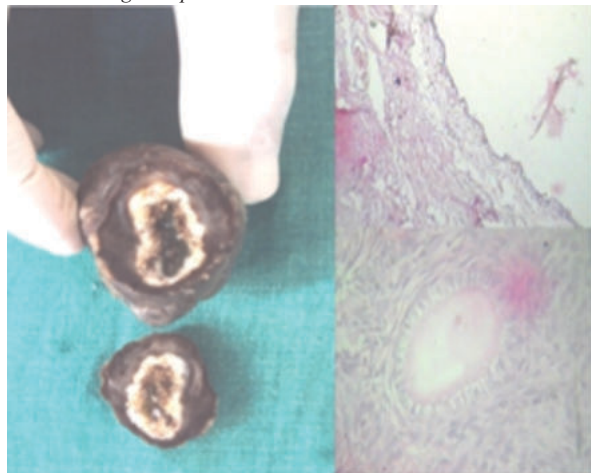


Figure 4: Gross And Histopathological Examination Of Lesion

DISCUSSION:

AOT itself is not a rare entity nowadays but there are only few AOT variants which arise from dentigerous cyst located in maxillary antrum. According to literature three variants exist of AOT on the basis of both clinical and radiological patterns. They are follicular type, which has a central lesion associated with an embedded tooth; the extrafollicular type, which has a central lesion and no connection with the tooth and the peripheral variety.

Amongst the central type, the follicular one initially is diagnosed as a dentigerous or follicular cyst with an impacted tooth. The extrafollicular type is usually present between, above or superimposed to the roots of erupted teeth. Mostly it is unilocular in nature. Peripheral type usually appears as a gingival swelling, may be palatally or lingually placed in relation to the erupted tooth. Uptill 2010, a systematic search on these type of lesions revealed that only seven such cases have been encountered, out of which only four cases occurred in maxillary sinus.⁷

Literature mentions amalgamation of features of multiple pathologies in AOT as a hybrid variant.^{8,9} Further a combination of both features of AOT and dentigerous cyst has been named as Hybrid variant of AOT,¹⁰ well visualized in our case. The reason for such hybrids is that both neoplastic as well as hamartomatous lesions can occur at any stage of odontogenesis process. Hence the odontogenic tumors like AOT with combined features of epithelial and mesenchymal components may arise within the odontogenic cyst. Similar pathologic changes may have been seen in our case as the patient gives history of progressive growth of swelling since 1 year.

As both AOT and dentigerous cyst are benign in nature, conservative management of such cases is possible. These encapsulated lesions can be surgically enucleated or curettage. As the recurrence rate of dentigerous cyst is less the treatment of choice is always enucleation. In very rare cases the aggressive behaviour of AOT has been seen, therefore the management of AOT includes complete removal of the tumor without any adjuvant modalities. Conclusively, we chose to enucleate the pathology in toto being the line of definitive treatment.

The origination of follicular type of variant of AOT mostly depends on the time of formation of lesion; before or after cystic expansion. If the origination occurs after the cystic expansion then it portrays its development from a dentigerous cyst as visualized in our case. But if the origination is before the cystic expansion then the lesion will be a solid tumour type of AOT. Therefore, 77% of follicular variant is often diagnosed as dentigerous cyst based on the clinical and radiographic findings as was experienced in our case.¹¹

In the present case, aspiration revealed straw coloured fluid indicating towards DC but on gross examination the soft tissue extended beyond the CEJ with a positive finding towards AOT. Furthermore, the cut section showed both the solid and cystic areas which was considered and named as a Hybrid variant of AOT.

CONCLUSION:

Very few cases of Hybrid variant have been reported till date where AOT collaborated with dentigerous cyst are present in the maxillary antrum. This thus highlights the importance of histopathological evaluation for cysts of jaw bones. Thus, meticulous histopathological evaluation is required from all enucleated cysts, which could contribute to the accurate diagnosis and appropriate management.

Further more research into the possibility of fourth type of a "hybrid variant" of AOT, apart from the already established three types of AOTs, is required.

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