



ADENOMATOID ODONTOGENIC TUMOR ASSOCIATED WITH A DENTIGEROUS CYST-A CASE REPORT

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ABSTRACT Odontogenic cysts and tumors are quite common occurrence in jaw bones although both of them occurring together are rare. Adenomatoid Odontogenic Tumor (AOT) is a benign odontogenic tumor arising from the complex system of dental lamina or its remnants. It is the fourth most common odontogenic tumor which is slow growing and has three variants – follicular, extrafollicular and peripheral. In this paper, we are reporting a rare case of AOT associated with Dentigerous cyst, where only few cases have reported previously.

KEYWORDS : Odontogenic tumors, Adenomatoid Odontogenic tumor, Dentigerous cyst, benign, dental lamina.

INTRODUCTION

Odontogenic tumors are the tumors of maxillofacial region which are derived from epithelial, ectomesenchymal, or mesenchymal components that are part of tooth-forming apparatus. The diagnostic criteria for Adenomatoid odontogenic tumor (AOT) have been known for more than 90 years. Earliest description was “**Epithelioma Adamantinum**” are from the works of Unal et al.¹ A variety of other terms has been used to describe this lesion, in which “**Adenoameloblastoma**” was common in use for many years because, the tumor was considered as histological variant of the solid/multicystic ameloblastoma.² In 1969, Philipsen and Birn presented a review based on 76 cases of AOTs that proved to be a distinct entity from the solid/multicystic ameloblastoma. They introduced the term “**Adenomatoid Odontogenic Tumor**” which was later adopted by the World Health Organization (WHO) in 1971³ which is now generally accepted nomenclature.

The 1992, WHO classification defines AOT as “a tumor of odontogenic 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. It is generally believed that the lesion is not a neoplasm.”

More than 95% cases of AOTs are intraosseous but extraosseous variants have also been documented. The tumor is twice more common in maxilla than mandible and have a strong predilection for anterior region. About three quarters of the cases occur in association with unerupted tooth in a pericoronal relationship, which led some authors to subclassify the tumor as follicular or extrafollicular variants. The follicular variant of AOT is often associated with an impacted tooth (about 77%)⁴ which is often diagnosed as Dentigerous cyst based on the clinical and radiographic findings.⁵ Very rarely, an AOT associated with an odontogenic cyst have been reported. Till date, only 14 cases have been reported. This article reports an unusual case of AOT associated with a dentigerous cyst around the crown of an unerupted maxillary canine.

CASE REPORT

A 19-year-old male patient came to the Department of Oral and Maxillofacial Surgery at Mahe Institute of Dental Science and Hospital with a complaint of missing tooth in relation to upper anterior

tooth region. Patient's previous medical and dental history was noncontributory.

However, patient was undergoing orthodontic treatment in the past 6 months. On general examination, patient was apparently normal. Extraoral examination did not show any facial asymmetry or regional lymphadenopathy. On inspection, a mild swelling was noticed in the region of 23 measuring about 1cm x 1 cm in dimensions. Mucosal surface of the alveolar ridge was even and no color change was noted. On palpation, it was non tender and bony hard in consistency. The patient was advised for an Orthopantomogram (OPG) for further investigation.

OPG shows, a well circumscribed, pericoronal radiolucency of an impacted 23 extending to the apical third of 22 and 24, along with displacement of 24.

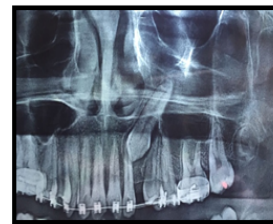


Fig1: OPG shows pericoronal radiolucency in relation to impacted 23

Based on clinical and radiographic examination, a differential diagnosis of **Adenomatoid Odontogenic Tumor or Dentigerous Cyst** was considered.

Surgical enucleation of the lesion was done under local anesthesia while canine was preserved.

The specimen was sent for histopathological examination. Orthodontic management was initiated for 23 to engage in functional occlusion.

Histopathology Features:

The H and E stained section shows a cystic lining and underlying connective tissue stroma. The cystic lining was 2-3 layers in thickness

resembling reduced enamel epithelium. Supporting connective tissue stroma comprises bundle of collagen fibers arranged in parallel to the lining epithelium. In focal areas, the stroma shows proliferating islands of spindle shaped epithelial cells and in other areas, epithelial cells were arranged in rosette pattern. Few tubular or duct-like structures with an inner layer of cuboidal cells were observed along with few small foci of calcification.

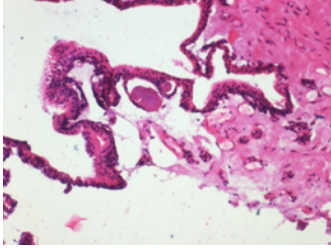


Fig2: Photomicrograph showing cystic lining of 2-3 cell layers thick (10x)

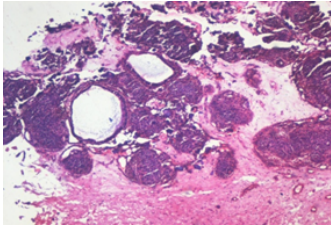


Fig3: Photomicrograph showing spindle shaped epithelial cells arranged in ductal pattern and rosette pattern (10x)

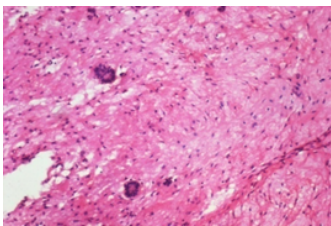


Fig 4: Photomicrograph showing odontogenic epithelial islands in connective tissue stroma (10x)

The histopathological features in correlation with clinical and radiographic features were suggestive of an **Adenomatoid odontogenic tumor associated with a Dentigerous cyst**

DISCUSSION

Adenomatoid Odontogenic Tumor (AOT) is a benign, non-neoplastic (hamartomatous) odontogenic tumor with a slow but progressive growth. The tumor occurs in tooth-bearing areas of the jaw and histomorphologically resemble to the components of dental organ.¹ Adenomatoid Odontogenic Tumor is also called two-third tumor, because two-third of the tumor occurs in maxilla, two-third occurs in young females, two-third of the cases are associated with the unerupted tooth.⁶ Sixty nine percent of AOTs are diagnosed in the second decade of life, and more than half of the cases (about 53%) occur during teenage years (mean age – 13years, range: 3 to 28 years of age). The female to male ratio for all age groups and variants of AOT together is very close to 2:¹

AOT is considered as fourth frequently occurring odontogenic tumor and it usually remain asymptomatic. The lesion is usually discovered on routine dental radiographic examination on account of unerupted or missing tooth. The tumor exhibits both intraosseous and peripheral forms.⁶ The intraosseous variant comprises a follicular and an extrafollicular type. The follicular variant shows a well-defined unilocular (round or ovoid) radiolucency associated with two-third of an unerupted tooth and found in between or superimposed on the roots of erupted permanent teeth. The extrafollicular variant is not associated with an unerupted tooth and it is well defined. The peripheral variant of AOT appears as gingival fibroma or epulis like growth attached to the labial or (more rarely) the palatal gingiva.¹

According to Philipsen et al., the pathogenesis of the tumor appears to derive from the dental lamina or its remnants which give rise to epithelial rests that proliferates in response to an unknown stimuli.¹

Due to the fact that the odontogenic tumors arise from the remnants of normal odontogenic apparatus it is difficult to unravel the relation between these two processes. The normal genetic regulation takes place through different signaling pathways, which includes four major families: Transforming Growth Factor [(TGF), which include Bone Morphogenic Proteins (BMPs)], Fibroblast Growth Factors (FGF), Hedgehogs (Hh), and Wingless (Wnt).⁸ Each family consists of several signals encoded by different genes. Alterations in these pathways of genes have been linked to a variety of developmental defects and there is evidence for tumor formation in adult life. Furthermore, p53 gene is a tumor suppressor gene and plays an important role in the regulation of cell proliferation. Mutation in the p53 gene yields a wild type p53 protein having a high cellular proliferation activity.⁸

The findings presented in this case correlates with the characteristic reports as presented in the literature, such as the lesion occurring in the maxillary region in association with an unerupted tooth. The radiographic findings are peculiar for Adenomatoid odontogenic tumor representing itself as a well-defined unilocular lesion, associated with crown of the unerupted tooth. Few cases of Adenomatoid odontogenic tumor associated with dentigerous cyst were also reported in the literature.

Early reports of Garcia-Pola et al.⁹ and Chen et al.¹⁰ describe the formation of an adenomatoid odontogenic tumor arising from dentigerous cyst. Histopathological findings of the cystic epithelial lining and the fibrous capsule in our case were in accordance to the description as given in the literature. Considering all the case reports of AOT associated with dentigerous cyst, it is much evident that the tumor is originating from the cystic lining. Moreover, WHO definition of AOT as given by Philipsen et al. supports the fact that cystic lining to be the progenitor for the tumor in many cases.¹¹ Since there is no report of its aggressive behavior or recurrence, this case was treated by conservative surgical enucleation.

CONCLUSION

Very few cases have been reported in literature where Adenomatoid odontogenic tumor is associated with a Dentigerous cyst. To evaluate this kind of tumor and its origin, further studies are needed to unravel the mechanism of tumor formation and progression. Since both the lesions are encapsulated, there are no reports of its aggressive behavior or recurrence. Further investigation in the gene regulation involved in the pathological development of AOT from dentigerous cyst, as well as the interrelationship among them provide a better insight into the pathology and may lead to the development of more efficient diagnosis, prevention and management.

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