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CASE REPORT

DENTIGEROUS CYST ASSOCIATED WITH ADENOMATOID ODONTOGENIC TUMOUR- A CASE REPORT

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ABSTRACT

Adenomatoid Odontogenic Tumour is a relatively uncommon benign odontogenic tumor which affects young individuals with a female predominance, mainly in the second decades of life. This lesion is most commonly affected in the anterior maxilla and usually associated with impacted tooth. In this paper, we present a case of extra-follicular AOT associated with dentigerous cyst affecting the mandibular canine-premolar region in a 25 year old female patient without any impacted tooth – a very rare situation.

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INTRODUCTION

Adenomatoid odontogenic tumor defined as "a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive changes in the connective tissue. The tumor may be partially cystic and in some cases solid lesion may be present as masses in the wall of large cyst. It is believed that lesion is not a neoplasm". AOT is an uncommon, benign, epithelial odontogenic tumor. Some authorities feel that, given the slow growth and circumscription of the lesion, it is best classified as a hamartoma rather than a true neoplasm (Simarpreet *et al.,* 2010). It was first described by Dreibaldt in 1907 as pseudoadenoameloblastoma (Singh *et al.,* 2012). Then it was described by Ghosh in 1934 as an adamantinoma of the maxilla and was first recognized as a distinct pathological identity by Staphne in 1948. Philipsen and Birn introduced the term *adenomatoid odontogenic tumor,* which was adopted by the World Health Organization (WHO) in 1971 (Kramer and Pindborg, 1971). AOT usually found in young patients, especially in the second and third decades of life, occurring predominantly in females with male: female ratio of 1:1.9.The lesion is benign (hamartomatous) and noninvasive with slow but progressive growth. It accounts for 2–3% of all odontogenic tumors (Shikha Gupta *et al.,* 2017).

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There are three variants of AOT, the follicular type (73%), extra follicular type (24%) and the peripheral type (3%). The follicular and extra follicular variants are intra osseous (central) and more commonly found in maxilla than in mandible at a ratio of approximately 2:1. The follicular variety usually associated with impacted tooth however the extra-follicular variant is not associated with the crown of an unerupted tooth. Instead, this variant presents as a well-defined, unilocular radiolucency between, above or superimposed upon the roots of erupted permanent teeth (Sumit Majumdar *et al.,* 2015). AOT is a tumor of odontogenic epithelium which are arranged in nodules or whorls of spindle-shaped or cuboidal epithelial cells. Around these highly cellular nodules, tumor cells form slender strands or cribriform pattern. However, these are clearly spherical microcysts and whatever the line of section, it always appear circular in form. They are lined by columnar cells which resemble ameloblasts and usually have their nuclei polarized away from the lumen. These microcysts may contain homogeneous eosinophilic material. Fragments of amorphous or crystalline calcification, which may be lamellated or sometimes resemble cementum, may be seen among the sheets of epithelial cells in the form of Liesegang ring. Occasionally, dentinoid or tubular dentine is also found (Sumit Majumdar *et al.,* 2015). The stroma is typically loose with scanty cellular elements, but contains thin-walled blood vessels, which are sometimes numerous.

Table 1. List of cases recorded so far-

Sl no	Reference	Age/sex	Race	Year	Site	Features
1	Valderrama	16/F	Philippino	1988	Maxilla	Unilocular radiolucency, surrounding tooth 14 crown
2	Warter et al	8/m	Nigerian	1990	Maxilla sinus	Unilocular radiolucency, surrounding tooth 13 crown
3	Tajima et al.,	15/m	Japanese	1992	Maxilla	A well defined radiopaque mass and crown of unerupted 28
4	Garcia-Pola et al.,	12/m	Spanish	1998	Maxilla	Unilocular radiolucency, surrounding tooth 23
5	Bravo et al.,	14/f	Not stated	2005	Maxilla	Unilocular radiolucency, surrounding tooth 23 crown
6	Nonaka et al.,	13/f	Brazil	2007	Maxilla	Unilocular radiolucency with few radiopaque areas 23 & 24
7	Chen et al.,	15/m	Chinese	2007	Maxilla	Impacted 23
8	Sandhu et al.,	25/f	Indian	2010	Maxilla	Impacted 13
9	J Baby John, Reena Rachel John	38/f	Indian	2010	Maxilla	Impacted 27
10	Khot and Vibhakar	17/f	Indian	2011	Maxilla	Impacted 33
11	Zarna Moosvi	13/f	Indian	2011	Mandible	Impacted 32
12	Vikramjeet Singh et al.,	15/f	Indian	2012	Maxilla	Impacted 13
13	Anshita Agrawal et al.,	15/f	Indian	2012	Maxilla	Impacted 23
14	Sushsruth Nayak et al.,	32/m	Indian	2012	Mandible	Impacted 43
15	Latti BR, Kalburge JV	15/f	Indian	2013	Mandible	Impacted 33
16	Anita DnyanobeMunde et al.,	20/f	Indian	2013	Mandible	Impacted 33
17	Harish Saluja et al.,	18/f	Indian	2013	Mandible	Impacted 43
18	Shivesh Acharya	14/f	Indian	2014	Maxilla	Impacted 13
19	Ludmila De Faro Valverde et al.,	17/f	Unknown	2014	Maxilla	Impacted 23
20	Sumit Majumdar et al.,	15/f	Indian	2014	Maxilla	Impacted 23
21	Shikha Gupta et al.	12/m	Indian	2015	Mandible	Impacted 33

Fig 1(A) Extra-oral clinical photograph of the patient, 1(B) Extra-oral photograph showing diffuse, uniform, firm, non tendered swelling involving the lower right third of the face. (1C) Intraoral photograph showing well defined, round, relatively large, soft to firm, fluctuant, non-tendered, non compressible, non pulsatile swelling involving lower right canine-premolar region in relation to muco-buccal fold

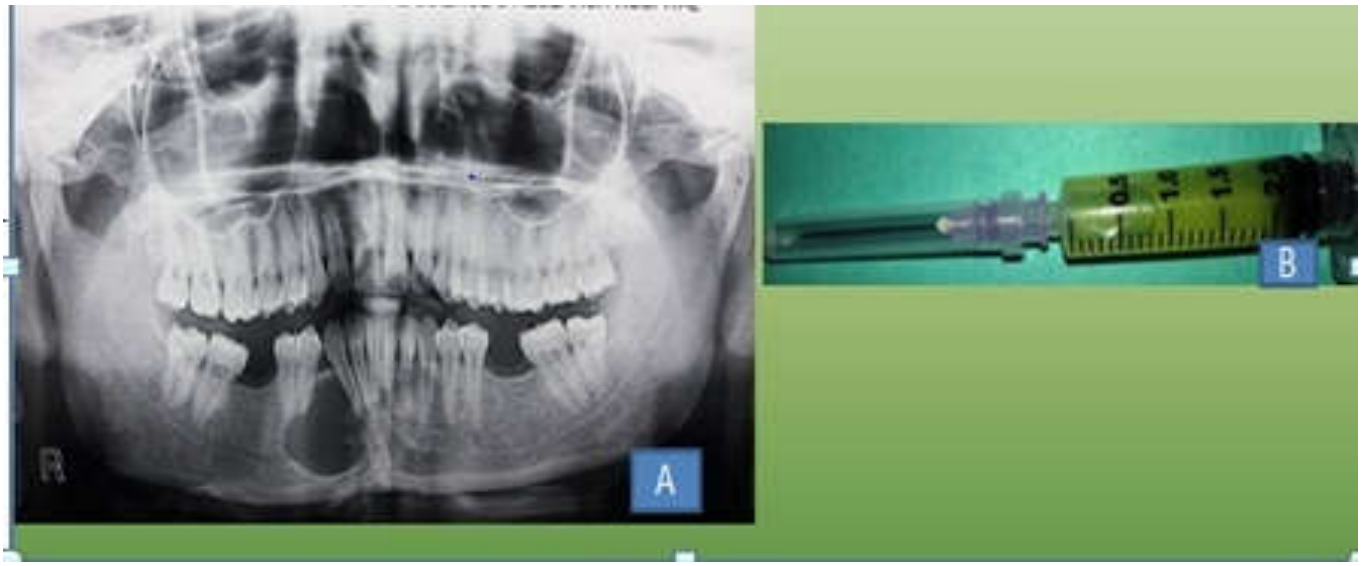

Fig 2(A) Orthopantomograph (OPG) of the jaws revealed a large, unilocular radiolucency involving the periradicular area of lower right lateral incisor extending up to the distal aspect of the root apex of right second premolar associated with displacement of regional teeth. 2(B) Clinical photograph showing aspirated straw coloured fluid

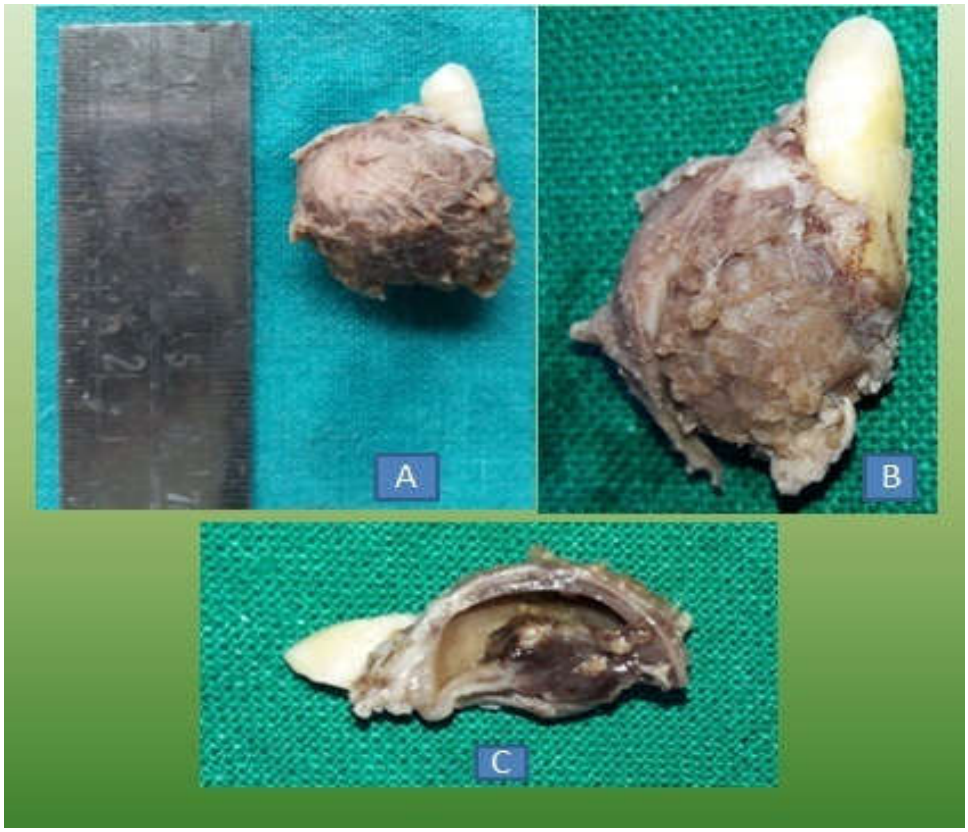

Fig 3(A,B,) On gross examination, the specimen was round(dimension 3cm x 3cm), grayish yellow in colour along with embedded canine, which was soft to firm in consistency.Fig(3C) Cut surface showing a cystic space with well defined fibrous capsule having solid mural proliferation

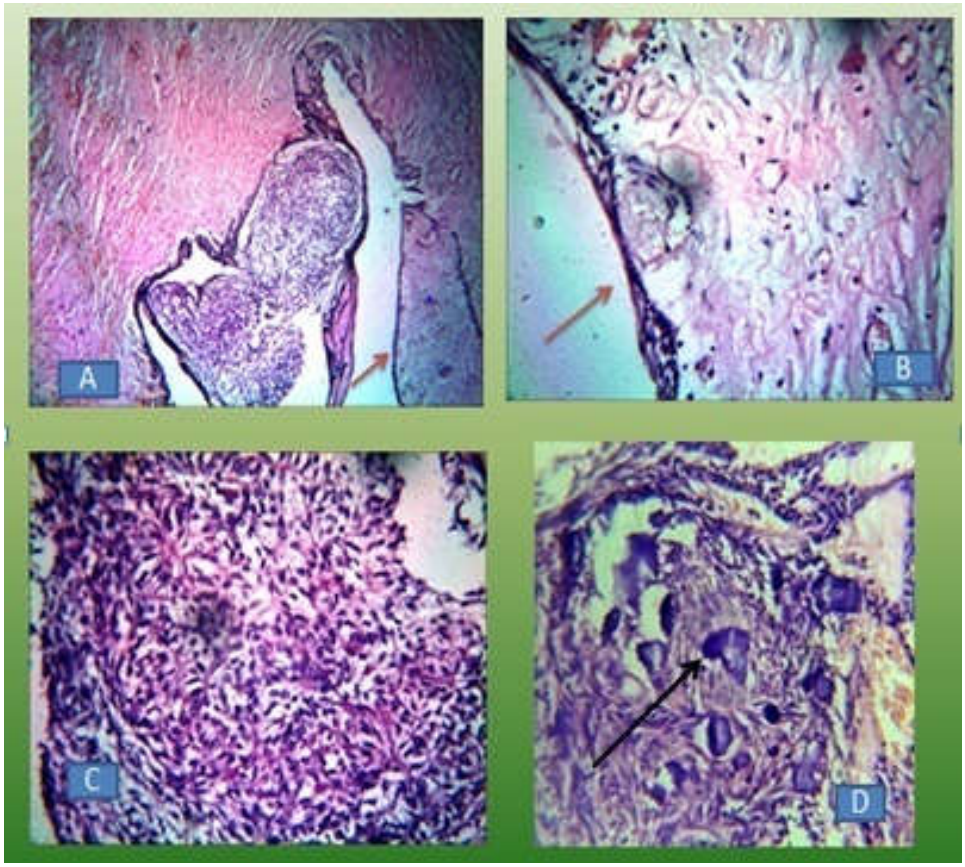

Fig (4A,B H&E 10X,40X) photomicrographs showing the presence of a thin cystic lining backed by fibro-vascular connective tissue stroma. Odontogenic epithelial cells proliferation were also noted in the cystic lining.. Fig (4C,D 40X) revealed odontogenic epithelial cells were proliferate in a rosette pattern. Areas of calcification (arrows) were also noted within the epithelial cells

Degeneration of the vessel walls, endothelium and perivascular connective tissue may be conspicuous. In some cases the solid lesion may be present only as masses in the wall of a large cyst. Most of them are resembles like that of dentigerous cyst. The epithelial lining of the odontogenic cyst may transform into an odontogenic neoplasm – like an ameloblastoma or AOT (Singh *et al.,* 2012). There have been very few reports of odontogenic cysts associated with Adenomatoid odontogenic tumors. The aim of this paper is to present a interesting case of AOT that originated in the wall of a dentigerous cyst.

CASE REPORT

A 25 years old female patient from semiurban area reported to the Department of Oral & Maxillofacial Pathology, Guru Nanak Institute of Dental Sciences and Research, Panihati, Kolkata with a chief complaint of swelling involving the lower right cheek region since last 2 month. The swelling was initially small in size which gradually enlarges and attained the present dimension over the time associated with mild discomfort. Medical and personal histories were non-contributory and the vital signs were within the normal limits. On extra-oral examination, there was a diffuse, uniform, firm to hard non tendered swelling involving the lower right third of the face which leads to the facial asymmetry(Fig-1A-B). The regional submandibular lymph nodes were palpable and nontendered- Overlying skin was free. Intraoral examination revealed the presence of a well defined relatively large, firm to hard, fluctuant, round to oval shaped, non-tendered, non compressible, non pulsatile swelling measuring about 3x1.5 cm, involving right lower canine-premolar region, associated with expansion of buccal cortical plates and mobility of regional teeth (Fig-1C). Radiologically, Orthopantomograph (OPG) of the jaws revealed a large, well defined, unilocular, well-corticated radiolucency involving the periradicular area of lower right lateral incisor extending up to the distal aspect of the root apex of left second premolar associated with displacement of the mandibular right canine and 1st premolar(Fig-2A).

Aspiration was performed from the lesion and about 2ml of straw-colored fluid was obtained(Fig-2B). On the basis of the clinical and radiographic findings, the differential diagnosis of Odontogenic Keratocyst, Dentigerous cyst, Unicystic Ameloblastoma, AOT was made. Then the patient was referred to the Oral & Maxillofacial Surgery department for further treatment & management. After that, the tumour mass was completely enucleated from the involved area and it was sent to our department for histopathological evaluation. On gross examination the specimen was round (3cm x 3cm), grayish-yellow in colour, firm, associated in relation to canine tooth. Cut surface of specimen showing a cystic space with well defined fibrous capsule having solid mural proliferation (Fig-3A-B). After H & E staining, the section revealed the presence of a cystic lining being composed of two to four layers of cuboidal cells, backed by fibro-vascular connective tissue stroma resembling primitive mesenchyme. Multiple islands of odontogenic epithelium, arranged in rosette, cords and ducts like patterns within the connective tissue stroma were also noted. The periphery of the duct like area was surrounded by tall columnar ameloblast like cells with palisaded nuclei. Presence of eosinophilic coagulum was also noted in the duct like spaces along with basophilic calcific deposits in the connective tissue (Fig-4A-B). The overall light microscopic features were suggestive of a hybrid neoplasm comprising of Dentigerous cyst and AOT.

DISCUSSION

AOT is a slow growing lesion which constitutes about 3% of all odontogenic tumor with a high predilection for anterior portion of maxilla-commonly associated with impacted canine among young females in the second decades of life (Shikha Gupta *et al.,* 2017). The case under discussion was a 25 years old female having a well defined swelling involving the anterior portion of mandible and presence of a relatively large, firm , fluctuant, round to oval shaped, non-tendered, non compressible, non pulsatile swelling measuring about 3x1.5 cm involving lower right canine-premolar region causing obvious facial deformity. According to the analysis of different case series the extrafollicular variant of AOT radiologically appears as a unilocular radiolucency found in between, above or superimposed on the roots of the erupted tooth and often resembles radicular, residual or lateral periodontal cysts (Simarpreet *et al.,* 2010). Our case also revealed the same radiographic feature. The case under discussion histopathologically revealed a cystic lining being composed of 2-4 layers of cuboidal cells backed by fibrovascular connective tissue resembling primitive mesenchyme. Multiple islands of odontogenic epithelium, arranged in rosette, ducts or cord like pattern within the connective tissue stroma were also noted. The periphery of the duct like areas were surrounded by tall columnar ameloblast like cells containing hyperchromatic nuclei situated away from the basement membrane mimicking reverse polarity. There is also presence of eosinophilic coagulum along with multiple foci of calcification in the connective tissue stroma. All these features are corroborative to the conventional histopathological finding of AOT except the cystic lining comprising of 2-4 layers of cuboidal cells suggesting the features of dentigerous cyst. According to the analysis of the different cases, AOT sometimes may be arising from the dentigerous cyst, represent a distinct hybrid variant which is completely isolated from the conventional variant of AOT as being described so far (Singh *et al.,* 2012). Very few cases have been reported that AOT may arise in association with DC. A systematic search of the English language medical literature revealed 21 such cases. Out of which 6 cases occurred in the mandible and all are associated with impacted tooth. 4 cases occurred in females of second decade (Table1).

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