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Case Report

Pathology Section

Adenomatoid Odontogenic Tumour Associated with Dentigerous Cyst: Case Report with Literature Review

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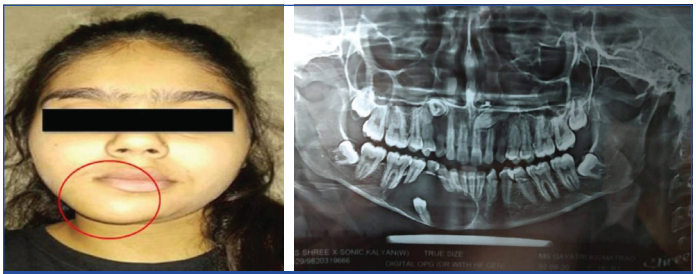
ABSTRACT

Adenomatoid Odontogenic Tumour (AOT) is a relatively uncommon benign odontogenic tumour composed of odontogenic epithelium in a variety of histoarchitectural patterns. Conventional AOT is predominantly seen in females in second decade of life exhibiting predilection for anterior region of maxilla. Very few cases of AOT associated with dentigerous cyst have been reported till date. The present case is unique associated with an impacted lower first premolar in the front region of the mandible of an 18-year-old female patient. The patient reported to the institute with gradually increasing swelling. The orthopantomogram revealed a unilocular radiolucency with displaced premolar. Histopathological examination confirmed 4×2.5×2 cm lesion in dimension, brownish black in colour, firm in consistency with a smooth surface grossly while cut surface revealed a cystic lumen with an impacted premolar embedded into the lining, cystic wall being nodular. Microscopic evaluation revealed a well encapsulated lesion with a thin 2-3 cell layered non keratinised stratified epithelial lining in patterns such as nests, rosette like structures and duct like structures. Thus the diagnosis of AOT arising from dentigerous cyst was confirmed. Patient's six month follow-up was uneventful. Literature search of similar cases with the review of hypothesised aetiopathogenesis is discussed in brief. The available data can help researchers resolve the uncertainty whether the AOT derived from dentigerous cyst could represent a distinct hybrid variety.

Keywords: Lower premolar, Mandible, Neoplastic transformation, Odontogenic cysts

CASE REPORT

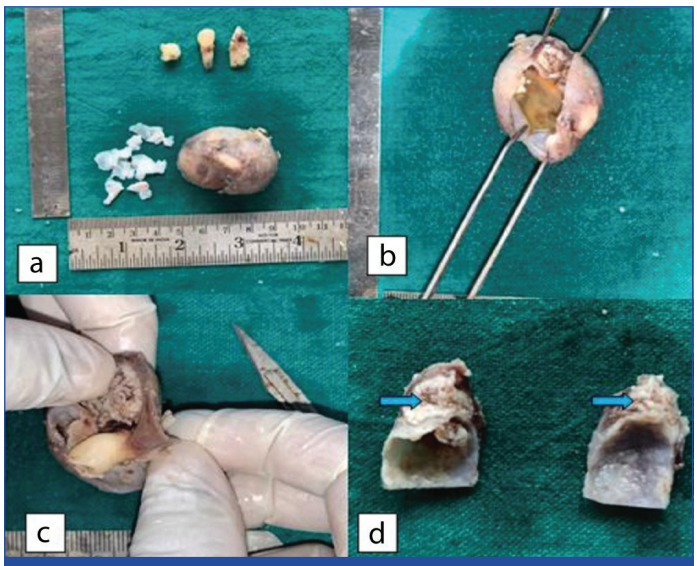
An 18-year-old female patient reported to Department of Oral and Maxillofacial surgery with a chief complaint of intra oral as well as extra oral swelling in the right back region of the lower jaw since 3-4 months resulting in a distinct facial asymmetry [Table/Fig-1]. Intra oral examination revealed a firm non tender swelling extending from 43 to 46 region. The overlying mucosa was non ulcerated with diffuse margins of the swelling. The orthopantomogram (OPG) demonstrated an ovoid unilocular radiolucency with well-defined borders [Table/Fig-2]. An unerupted premolar within the radiolucent lesion was seen displaced towards the lower border of the mandible. The deciduous molars were over retained. On the basis of clinical and radiographic findings, a provisional diagnosis of dentigerous cyst was made with a differential diagnosis of ameloblastoma and AOT.



[Table/Fig-1]: Facial asymmetry with swelling on lower left front region; **[Table/Fig-2]:** Orthopantogram showing unilocular well defined radiolucency with corticated borders embedding impacted 1st premolar of right-side in mandible. (Images from left to right)

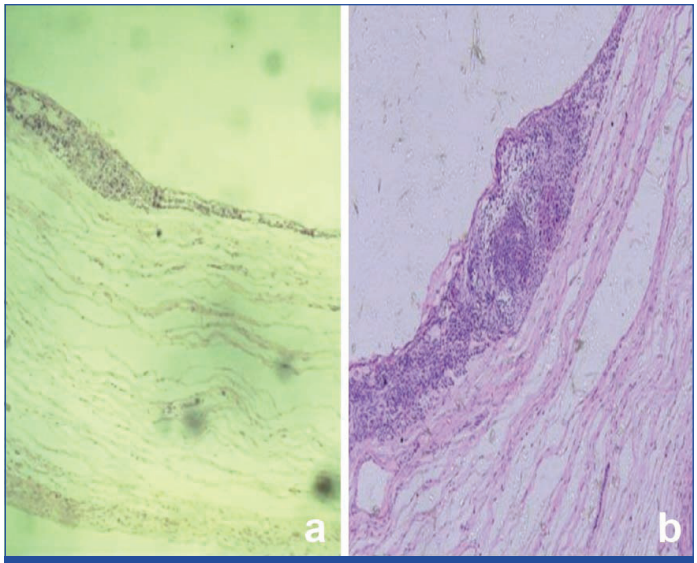
Enucleation of the lesion was planned under local anaesthesia. Before surgery, preoperative arch bar was secured in maxillary and mandibular arches. Crevicular incision was given from 32 to 46. The mucoperiosteum was separated from the tumour lining. The tumour was curated from the surrounding bone in toto along with the embedded premolar and the specimen was submitted for the histopathological examination.

On gross examination, the tissue specimen was 4×2.5×2 cm in dimension, brownish black in colour, firm in consistency with a smooth surface. The cut surface revealed a cystic lumen with an impacted premolar embedded into the lining. The cystic wall also demonstrated nodular thickening in some areas [Table/Fig-3].

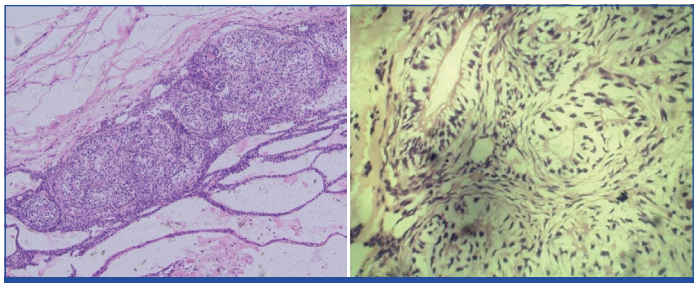


[Table/Fig-3]: Gross examination of the biopsy specimen: a) Enucleated tissue in toto with extracted deciduous teeth and bone fragments; b and c) Impacted tooth in the cystic lining of the tissue; d) specimen cut into two sections with cystic wall showing proliferated tissue (arrow).

Microscopically, Haematoxylin and Eosin (H&E) stained section showed a well encapsulated lesion with a thin 2-3 cell layered non keratinised stratified epithelial lining [Table/Fig-4]. Focal areas of intra luminal proliferation of spindle shaped cells in patterns such as nests, rosette like structures and duct like structures were noted [Table/Fig-5,6]. These duct like areas were lined by a single layer of cuboidal or columnar epithelial cells. Anastomosing cords of basaloid epithelial cells were arranged in plexiform configuration extending between the cell rich nodules. Irregular to round calcified bodies and droplets of eosinophilic hyaline material were observed among the cellular



[Table/Fig-4a,b]: (Photomicrograph of Haematoxylin and Eosin (H&E) stain) scanners view 4X magnification showing lining of cystic wall depicting reduced enamel epithelium suggestive of dentigerous cyst with neoplastic transformation.



[Table/Fig-5]: (Photomicrograph Haematoxylin and Eosin (H&E) stain) Low power: 10X magnification showing Rosette formation and ductal pattern of epithelial cells; **[Table/Fig-6]:** Photomicrograph (H&E stain) High: 40X magnification showing duct like structures, lined by a single layer of cuboidal and columnar epithelial cells along with rosette formation. (Images from left to right)

proliferations. Based on these features, histopathological diagnosis of AOT arising from dentigerous cyst lining was made. Six month follow-up of the patient was uneventful.

DISCUSSION

The AOT is a relatively rare benign hamartomatous slow growing epithelial lesion of odontogenic origin accounting for 2-7% of odontogenic tumours [1,2]. The lesion is also known as a two-third tumour since two-third cases are reported in young individuals, two-third cases have female predilection, two-third of the cases are associated with impacted maxillary canine [3-7]. The lesion was first described by Ghosh in 1934, since then many AOT cases have been identified with impacted teeth. AOT is normally associated with follicular tissue of unerupted teeth confirming dental germ tissue origin [2]. The tumour maybe partly cystic or may occur as a solid lesion. There are three variants of AOT:

- Intraosseous or central AOT which can be
- Follicular type- associated with crown of an impacted tooth
- Extrafollicular type- not associated with an impacted tooth
- Peripheral AOT- presents as gingival swelling

It has been reported that some odontogenic cysts occur in association with odontogenic tumours [8]. Reports of AOT associated with different types of cysts and odontogenic neoplasms are present in the literature [9]. An extensive review of 500 cases of AOT has been conducted by Philipsen HP et al., [10]. He postulated that the follicular type of AOT develops from nests of cells within the dental lamina and therefore surrounds the tooth [10]. Very few cases of AOT associated with dentigerous cyst have been reported so far [1,3,4,6-8,11-26].

A systematic search of the English language medical literature revealed only 23 such cases. A review of these cases is mentioned in tabular form [Table/Fig-7] [1,3,4,6-8,11-26].

The present case is unique as it is only fourth of its kind occurring in posterior mandible according to the reports in the literature.

Author and Year	Age/Sex	Clinical features	Radiographic examination
Water A et al., 1990 [11]	8/M	Swelling with tenderness in right cheek region with impacted canine and premolar	Well circumscribed radiolucent lesion
Tajima Y et al., 1992 [12]	15/M	Lesion associated with left maxillary sinus included impacted 3rd molar	Well defined radiopaque mass
Takahashi K et al., 2001 [13]	22/M	Swelling and painless nasal obstruction in left cheek and nasal cavity with impacted 3rd molar	Large lesion with expansion of sinus and thinning of bony sinus wall
Bravo M et al., 2005 [14]	14/M	Swelling and sharp pain in left cheek region with impacted canine	Expansile lesion with sclerotic rim
Nonaka CF et al., 2007 [15]	13/F	Mass in maxillary region associated with impacted canine	Circumscribed radiolucent lesion with few radiopaque areas
Chen YK et al., 2007 [16]	15/M	Swelling and parasthesia or anaesthesia in maxillary left side with impacted canine	Well defined unilocular radiolucency
Sandhu SV et al., 2010 [17]	25/F	Swelling and nasal obstruction in left maxilla with unerupted canine	Large lesion on CT scan
John JB and John RR, 2010 [18]	39/F	Pain and swelling in left upper jaw region with impacted 2nd molar	Well defined, unilocular, corticated radiolucency
Moosvi Z et al., 2011 [19]	13/F	Swelling in chin region with associated unerupted lateral incisor	Well defined radiolucency with sclerotic borders
Agrawal A et al., 2012 [7]	15/ F	Swelling in left maxilla with impacted canine	Well defined unilocular radiolucency
Singh V et al., 2012 [20]	28/F	Swelling, dull and intermittent pain in maxillary right buccal region with associated impacted canine. other symptoms include palpable lymph nodes	Well defined radiolucency with corticated borders
Munde AD et al., 2013 [21]	20/F	Swelling in anterior mandible associated with uneruptedcanine	Well defined unilocular radiolucency with corticated borders. External root resorption of adjacent tooth
Saluja H et al., 2013 [22]	18/F	Swelling in anterior region of lower jaw with impacted canine	Well defined corticated radiolucency
Rathod V et al., 2014 [3]	13/M	Swelling in mandibular left anterior associated with impacted 1st premolar	Unilocular radiolucency with corticated borders
Acharya S et al., 2014 [6]	14/M	Swelling and pain on right-side of face with unreuptedcanine	Well defined radiolucency with displacement of other teeth
Geetha NT et al., 2014 [23]	14/M	Swelling in left middle third of face with uneruptedcanine	Well circumscribed, inverted pear shaped radiolucent lesion
Manjunath BS et al., 2015 [1]	20/ F	Swelling and mild pain in hard palate region with impacted canine	Unilocular radiolucency
Manjunath BS et al., 2015 [4]	19/F	Swelling, discomfort and dull intermittent pain in mandibular left posterior region with impacted 1st premolar	Unilocular radiolucency with displacement of teeth
Kalburge JV et al., 2015 [24]	Case 1: 18/F Case 2: 15/F	1: Swelling associated with canine impaction 2: Swelling with impacted canine	Unilocular radiolucency
Gupta S et al., 2016 [25]	12/M	Swelling, dull intermittent pain, and lymphadenopathy in anterior mandible with impacted canine	Well defined unilocular radiolucency, displacement of teeth and external root resorption of adjacent teeth
Nath S et al., 2017 [26]	13/F	Swelling associated with maxillary left side with impacted canine	Unilocular radiolucency with corticated borders

Patel HB et al., 2020 [8]	15/M	Swelling in mandibular anterior region with impacted canine	Well defined radiolucency with displacement of other teeth
Present study; 2021	18/F	Swelling in right mandibular region with impacted Premolar	Unilocular well-defined radiolucency with embedding impacted 1st premolar of right-side in mandible and also over-retained deciduous molars

[Table/Fig-7]: Similar cases of AOT with dentigerous cyst reported in literature [1,3,4,6-8,11-26].
AOT: Adenomatoid odontogenic tumour; CT: Computed tomography; M: Male; F: Female; *Provisional Diagnosis for all the cases was given as Dentigerous cyst except Gupta S et al., in 2016, clinically given as central giant cell granuloma

The age range of patients reported in this review was from 8-39 years with the average age being 20 years i.e., second decade of life. This age range correlated with the conventional AOT cases. AOT associated with dentigerous cyst is slightly more common in females than in males. Predominant chief complaint of the patients was the long-standing swelling progressing over few months while few cases additionally reported dull intermittent pain and slight discomfort. Along with these common symptoms, nasal obstruction, symptomatic lymph nodes and paraesthesia of the jaw were other symptoms [16,17,20,25]. Maxillary predilection in anterior region is much higher in conventional AOT but not so much in AOT associated with dentigerous cyst. Canine is the most common impacted tooth associated with the lesion with only two exceptions of association with premolar [3,4]. The case in this study presented with a swelling in association with an impacted mandibular premolar.

When associated with dentigerous cyst AOT may demonstrate grossly and microscopically one or more cystic cavities lined by non keratinised stratified squamous epithelium. The tumour portion in the current case was continuous with the cyst lining. Considering this, we believe that a dentigerous cyst with unerupted tooth develops first and through a stimulus yet to be determined; AOT would subsequently arise from the epithelial rests of dental lamina within the odontogenic cyst lining. These observations were supported by other studies by Manjunatha BS et al., and Sandhu SV et al., who believe that the dentigerous cyst undergoes neoplastic transformation [4,17]. However, Agrawal A et al., and Chen YK et al., still consider the possibility of such lesion representing a distinct hybrid variant [7,16].

CONCLUSION(S)

The AOT and dentigerous cyst are benign encapsulated lesions, hence conservative surgical enucleation or curettage is a treatment of choice. There have been no reports of aggressive behaviour of AOT. Recurrences are very rare after complete removal of the lesion. In our case the postoperative course was uneventful without any signs of recurrence so far.

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