

RELATIONSHIP BETWEEN DENTIGEROUS CYST AND AOT: A CASE REPORT AND RIVIEW OF LITERATURE OF PAEDIATRIC CASES

Case Report

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ABSTRACT

Adenomatoid odontogenic tumour (AOT) is an epithelial tumor with an inductive effect of odontogenic ectomesenchyme. This slowly growing tumour, also considered as a hamartoma is more common in females. The lesion usually associated with the crown of an enclosed tooth, most commonly the maxillary canine. The current WHO classification of odontogenic tumour defines AOT as being composed of the odontogenic epithelium in a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma and characterized by slow, but progressive growth. It has a relative frequency of 2.2 - 13% and often misdiagnosed as an odontogenic cyst on clinical examination. In this paper we present a case of Adenomatoid odontogenic tumour associated with a dentigerous cyst affecting the left maxilla in a 17 year old male. We also discussed clinical, radiographic, histopathologic and therapeutic features of the case with a post-operative follow up to see their recurrence occurs or not.

Key Words: Adenomatoid odontogenic tumour (AOT), benign oral tumour, anterior maxillary tumour, hamartoma, Cemento-enamel junction (CEJ)

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INTRODUCTION

AOT is usually a symptomatic slow grow lesion. It was first discovered by Steenland in 1905 (Steenland 1905). [1] In WHO classification of 2005 AOT is included under odontogenic epithelium with mature, fibrous stroma without odontogenic ectomesenchyme. [2]

The age range between which AOT occurs varies between 3 and 82 years. More than two-thirds are diagnosed in the second decade of life and 90% are found before the age of 30. More than half of the cases occur among teenagers. The male: female ratio is 1:1.9. [3,4]

Rick has reported AOT to occur with many types of cysts and neoplasm including dentigerous cyst, calcifying odontogenic cyst, odontoma and ameloblastoma. In relation with a dentigerous cyst the AOT may demonstrate, grossly and microscopically, one or more associated cystic cavities. Some of these cysts are lined by nonkeratinized stratified squamous epithelium which is similar to the lining of the dentigerous cyst or lined by less structured membrane that may demonstrate bud-like extensions into the connective tissue. [5]

Intraosseous AOT may be found in association with unerupted permanent teeth (follicular type), in particular the four canines that account for 60% with the maxillary canines alone accounting for 40%. [6]

CASE PRESENTATION:

A 17 year old male patient came to the department of Oral & Maxillofacial Surgery with a chief complaint of swelling in his left upper jaw since last 6 months. On examination we found a diffuse non-tender swelling present in left anterolateral part of maxilla and there was expansion of buccal cortical plate and palatal bone (Figure 1).

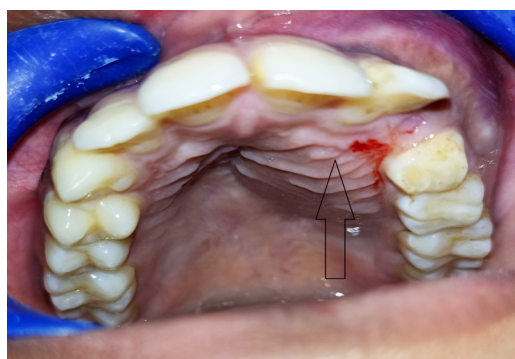


Figure 1: Pre-operative intraoral photo with swelling in the left maxilla region with missing 23, 24, 25

Initially the swelling was small and it gradually increased in size. Overlying mucosa appeared to be normal. Over-retained deciduous teeth 64, 65 seen and missing teeth were 23, 24 & 25. On aspiration we found yellowish straw coloured fluid. On radiographic examination a unilocular radiolucency seen in anterior maxillary region extending from pyriform aperture to posterior-laterally towards the maxillary buttress region. Superior it extends from infra-orbital margin to inferiorly till the alveolar process. Impacted teeth 23, 24 & 25 were also seen associated with the radiolucency suggestive of a dentigerous cyst. (Figure 2)

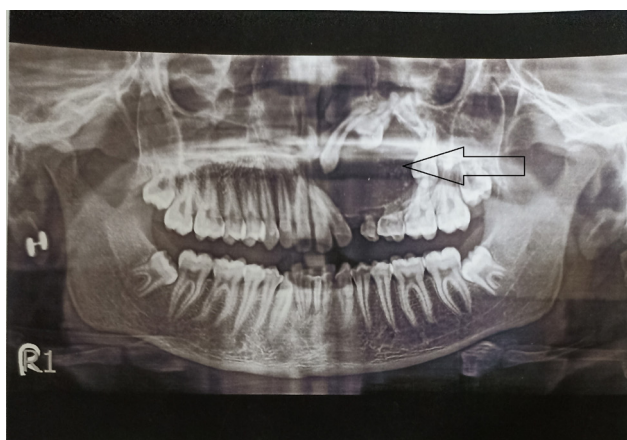


Figure 2: In OPG (Orthopantomogram) unilocular well defined radiolucent cyst seen in left anterior maxillary region

Axial CT scan finding showed an osteolytic heterogeneous cystic lesion occupying the left anterolateral maxilla of size (5cm x6cm) with expansion of buccal and palatal cortical plate. The coronal scan showed an impacted teeth associated with the cystic cavity. The cystic lesion encircled the crown of tooth displaced to infra orbital margin giving an appearance of a dentigerous cyst. (Figure 3)

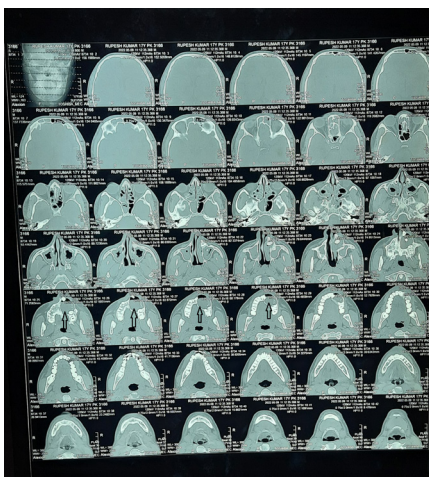


Figure 3: Axial CT scan finding showed an osteolytic heterogeneous cystic lesion occupying the left anterolateral maxilla of size (5cm x6cm) appx. with expansion of buccal and palatal cortical plate

TREATMENT PLAN:

Based on the clinical and radio graphical findings the lesion was diagnosed as dentigerous cyst. Enucleation of the tumor under general anaesthesia was selected as the treatment plan. After induction of general anaesthesia with oral intubation a trapezoidal incision was placed extending from left maxillary lateral incisor to the left maxillary 2nd molar. A trapezoidal mucoperiosteal flap raised to expose the tumor. (Figure 4)

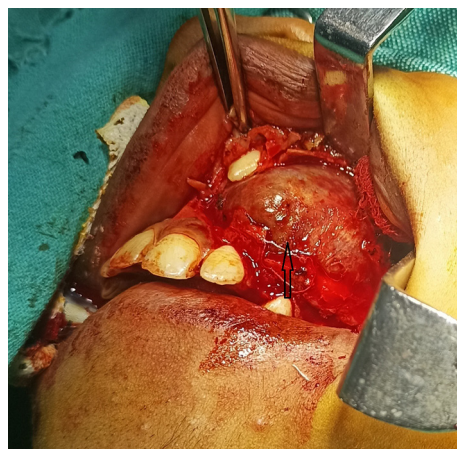


Figure 4: A trapezoidal mucoperiosteal flap raised to expose the tumor.

The thin labial cortical plate was removed cautiously with the help of a bone ronger without disrupting the cystic lining. Complete decortication of the over line labial cortex was done to expose the underlying cystic lesion. Careful enucleation of the cyst was done along with removal of the attached canine tooth (23). The impacted teeth 23 & 25 were also removed. (Figure 5) Meticulous debridement of the surgical site was done with closure of the wound by 3.0 resorbable polyglactin suture was performed.

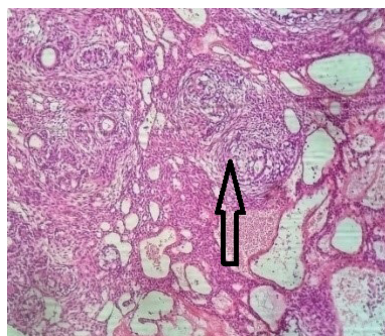


Figure 5: Surgical gross revealing a cystic lesion with embedded 23, 25

Gross examination of the specimen of size 4.4 cm showed a thick cystic lining surrounding the crown of the tooth 24 and attached to the cemento enamel junction (CEJ). The surgical specimen was sent for histopathological examination. The histopathological examination showed in H&E stained section reveals fibrocellular connective tissue stroma consisting predominantly of spindle shaped epithelial cells arranged in duct and rosette like pattern, with central empty space. (Figuer 6) These cells surrounding the spaces are tall columnar with reversal of polarity of nuclei away from lumen. (Figuer 7) The surrounding capsule is fibrocellular with bundles of collagen fibres, fibroblasts and mild inflammation. (Figuer 8)



Figuer 6: H&E stained section reveals fibrocellular connective tissue stroma consisting predominantly of spindle shaped epithelial cells arranged in duct and rosette like pattern, with central empty space. (10X)



Figuer 7: cells surrounding the spaces are tall columnar with reversal of polarity of nuclei away from lumen (10x)



Figure 8: Thin anastomosing strands of basaloid cells in plexiform pattern arranged. (10x)

DISCUSSION:

Adenomatoid odontogenic tumor (AOT) is a benign, non-invasive lesion comprising of approximately 3% of all odontogenic tumors ranking behind odontomes, cementoblastomas, myxomas and ameloblastomas. [7] Adenomatoid odontogenic tumor has been reported in patients from 3 to 82 years of age, its predilection for young patients is established in literature, 69% cases are diagnosed between the ages of 10 and 19 years. Pericoronal (dentigerous, follicular) Adenomatoid odontogenic tumors are diagnosed at an earlier age than other variants. [8] AOT frequently resembles lesions like dentigerous cyst or ameloblastoma. In-fact, 77% of follicular type AOT is initially diagnosed as dentigerous cysts. [9]

Gadewar and Srikant reported a case of AOT having a large cystic space lined by thick stratified squamous epithelium and suggested that the presence of AOT in the nodules of cystic lining gives a histological proof that AOT had transformed from a cyst. [10] Although AOT is called the perfect imitator of the dentigerous cyst, the characteristic clinical features renders the diagnosis relatively obvious. Reports have also shown the size of the lesion to be as large as 12 cm. [11] It has a decisive sex predilection for females, while few cases have also been seen in males. [12] In our case AOT and dentigerous cyst histological features were found in same tumor. The clinical, radiographic features were consistent with findings of dentigerous cyst. AOT is usually solid but can arise along with dentigerous cyst. We have enumerated cases in **table-1** were AOT has arisen along with dentigerous cyst in paediatric age group.

The structure of the cyst and its insertion around the crown of an unerupted tooth were typical of a dentigerous cyst. Odontogenesis is a complex process and neoplastic or hamartomatous lesion can occur at any stage of Odontogenesis. The secondary development of an ameloblastic proliferation, whether hyperplastic or neoplastic, is well known but remains controversial.

In this case, the multifocal cellular proliferation had the structure of an AOT. Its mural development in a dentigerous cyst is not uncommon. The tumor is well encapsulated and shows an identical benign behaviour. Therefore, conservative surgical enucleation produces excellent outcome without recurrence. [7] (Figuer 9)

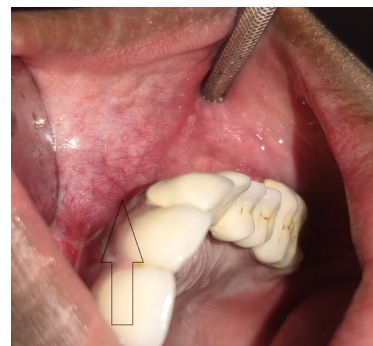


Figure 9: 6 months of follow up showed no recurrence.

TABLE 1: A systemic Clinical data of the reported cases of Adenomatoid odontogenic tumor arising from a dentigerous cyst in paediatric age group till date

Authors	Age/Sex	Race	Site	X-Ray finding	Other finding
1. Valderrama[7]	16/F	Philippians	Maxilla	Unilocular radiolucency, surrounding tooth 14 crown	Presence of complex odontoma
2. Water et al. [8]	8/M	Nigerian	Maxilla	Unilocular radiolucency, surrounding 13 crown	Contained melanocytes and melanin-laden epithelial cells
3. Tajima et al.[9]	15/M	Japanese	Maxillary antrum	A well-defined radiopaque mass and crown of unerupted 28	-----
4. Garcia-Pola Vallejo et al. [10]	12/M	Spanish	Maxilla	Unilocular radiolucency, surrounding tooth 23 crown	Agensis of tooth 15 and 24
5. Bravo et al.[11]	14/M	Notstated	Maxilla	Unilocular radiolucency, surrounding tooth 23 crown	Expanding to sinus
6. Nonaka et al.[12]	13/F	Brazil	Maxilla	Unilocular radiolucency with few radiopaque areas 23 and 24	-----
7. Chen et al. [13]	15/M	Tiwan	Maxilla	Radiolucency around upper deciduous canine	Odontoma-like areas were observed
8. Khot and Vibhakar [14]	17/F	Indian	Mandible	Unilocular radiolucency surrounding unerupted 33	Involving whole ramus with pus discharge
9. Agarwal et al. [6]	15/F	Indian	Maxilla	Unilocular radiolucency, tooth 23 crown surrounded	Root resorption of adjacent teeth
10. Vimi S Mutalik et al. [15]	14/F	Indian	Maxilla	Unilocular radiolucency with impacted 23	Root resorption of 63
11. Our Case	17/M	Indian	Maxilla	Unilocular radiolucency with impacted 23	Over-retained 64.65

CONCLUSION:

Such tumors which have hybrid features of AOT and Dentigerous cyst creat confusion regarding the right nomenclature and diagnosis of this hybrid tumors.

DECLARATION OF CONFLICTING INTERESTS:

The authors declared no potential conflicts of interest with respect to the research, authorship and publication of this article.

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