

## ADENOMATOID ODONTOGENIC TUMOR – A CASE REPORT

## Oral Pathology

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## ABSTRACT

AOT is a benign epithelial odontogenic tumor of jaw bone. It constitutes about 2.2 – 7.1% of the odontogenic tumors. Clinically and radiographically AOT should always be considered in the differential diagnosis of lesions in Anterior maxilla. Histopathology remains the gold standard in diagnosing these lesions. Enucleation and curettage is the most common treatment modality. We present case report of an Adenomatoid odontogenic tumor in a 13 year old female patient .

## KEYWORDS

adenomatoid odontogenic tumor , 2/3<sup>rd</sup> tumor , Anterior maxilla , impacted canine , young female

## INTRODUCTION –

Adenomatoid odontogenic tumor (AOT) is an uncommon, hamartomatous, benign, epithelial lesion of odontogenic origin.<sup>1</sup> It has been considered as a hamartoma rather than a true neoplasm because of its limited size, minimal growth potential and the lack of recurrence.<sup>5</sup> Adenomatoid odontogenic tumor is commonly known as 'two-thirds tumor,' as 2/3<sup>rd</sup> of adenomatoid tumors occur in the maxilla, 2/3<sup>rd</sup> occur in young females, two-thirds are associated with un-erupted teeth and two-thirds of the affected teeth are canines.<sup>1</sup>

## CASE REPORT -

A 13 year old female patient reported to the Dept of Dentistry , IGIMS with the chief complaint of swelling involving the left side of the face since 2 years. The medical and dental history were non-contributory . On extra oral examination , a swelling was seen on the left mid facial region extending anteroposteriorly from midline till outer canthus of the eye and superoinferiorly from lower eyelid till corner of the mouth measuring 5\*4 cms. ( Fig-1) On palpation the swelling was non tender , firm in consistency with ill defined margins. On intra-oral examination , swelling was seen extending from incisor till canine region obliterating the labial mucosa. (Fig-2) Based on the history and clinical examination , a provisional diagnosis of benign odontogenic tumor of left maxilla was suggested. Patient was then sent for further radiological investigations. The orthopantomography revealed a well defined unilocular radiolucency with radiopaque border measuring 3 \*3 cm surrounding the crown of the left maxillary impacted canine.(Fig-3) In order to determine the extent of the lesion , patient was then advised for CT Scan of face (Fig- 4). Subsequently surgical enucleation of the lesion was performed and the tissue along with the attached tooth was sent for histopathological examination.(Fig-5) Grossly the specimen was well encapsulated , firm and greyish black in colour measuring 3\*1.8\*1.5 cm. The histopathology of the tissue showed odontogenic epithelial cells forming multiple solid nodules of varying sizes with scanty connective tissue stroma. Few areas within these nodules demonstrated duct like spaces. (Fig- 6)The ductal lining was lined by columnar cells with nucleus placed away from the central lumen. The lumen was filled with eosinophilic material resembling amyloid. Few odontogenic cells surrounding the tumor nodules consisted of stellate reticulum like spindle shaped cells arranged in sheets.(Fig- 7) Irregular , spheroidal and globular masses of calcification were seen within the epithelial and the connective tissue component. (Fig- 8) Connective tissue was fibrous with areas of hyalinization and marked vascularity. Based on the above features , a confirmatory diagnosis of Adenomatoid odontogenic tumor was rendered following which complete surgical excision of the tumor under general anaesthesia was planned.



Fig -1

Fig -2



Fig-3

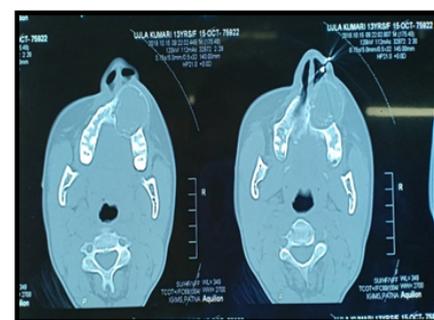


Fig-4



Fig-5

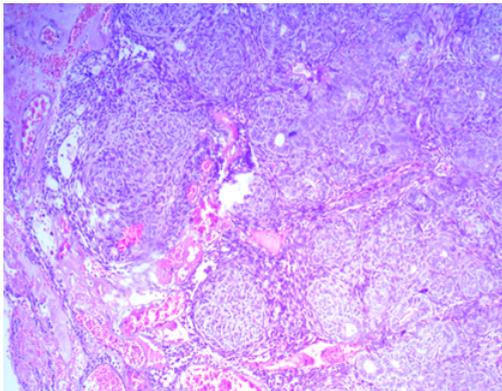


Fig-6

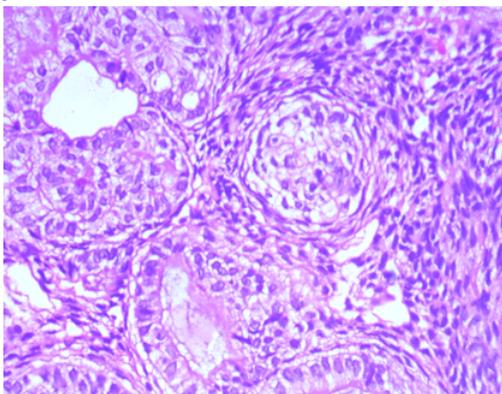


Fig-7

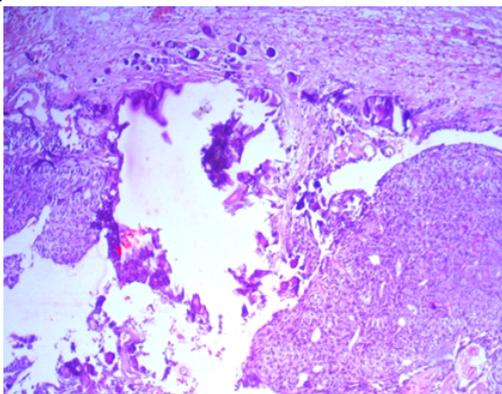


Fig-8

**DISCUSSION-**

It was first described by Driebaldt in 1907, as a pseudo-adenoameloblastoma.<sup>1</sup> Cystic presentation of AOT has been reported way back in 1915 by Harbitz who reported the lesion as “cystic adamantoma”<sup>4</sup> Unal et al<sup>10</sup> produced a list containing all nomenclatures for AOT reported in the literatures. Many different names like adenoameloblastoma, ameloblastic adenomatoid tumour,

epithelioma, adamantinum or teratomatous odontoma have been used before to define the lesion currently called AOT. Philipsen and Birn (1971) proposed the widely accepted and currently used name, adenomatoid odontogenic tumor.<sup>1</sup> The current World Health Organisation (WHO) classification of odontogenic tumors defines AOT as being composed of the odontogenic epithelium in a variety of histoarchitectural patterns, embedded in mature connective tissue stroma, and characterized by slow, but progressive growth.<sup>1</sup> The origin of AOT is believed to be from an odontogenic source; the cytologic features are similar to those of the enamel organ, dental lamina, reduced enamel epithelium, and / or their remnants.<sup>11</sup> Regarding its pathogenesis, the lesion originates from odontogenic epithelium (enamel organ or dental lamina remnants) with inductive influence on odontogenic ectomesenchyme and consequent production of dentinoid material.<sup>9</sup> AOT shows centrifugal expansion (uniform expansion in all directions). It has been hypothesized that at an early stage AOT may expand the cortical plates, which within the cancellous bone spread linearly and then later may affect the cortical plates by expansion/resorption.<sup>1</sup>

There are three clinicopathological variants of AOT, namely intraosseous follicular, intraosseous extrafollicular and peripheral.<sup>5</sup> The follicular type accounts for about 73% of all AOTs. The extrafollicular variant for about 24% of all AOTs<sup>1</sup>. and the peripheral variant for about 3% of all AOTs<sup>6</sup>. The follicular type shows a well defined unilocular radiolucency associated with the crown and often part of a root of an unerupted tooth, thus mimicking a dentigerous or follicular cyst. However the extrafollicular type is not associated with an unerupted tooth<sup>7</sup>. According to Philipsen, the extrafollicular type can be found in four different locations: intra-osseous and unrelated to retained teeth, intra-radicular, periapical and in the middle third of the root<sup>7</sup>. The peripheral variant occurs primarily in the gingival tissue of tooth-bearing areas<sup>5</sup>. Radiographically AOT often involves the crown, expansion of the tumor causes displacement of adjacent teeth, and tooth displacement is more common than root resorption<sup>8</sup>. Minute radiopacities (califications) around the retained tooth are seen and considered characteristic, but not pathognomonic.<sup>1</sup> Radiological findings of AOT simulate many other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts or tumor, ameloblastoma, keratocystic odontogenic tumor. The above-mentioned lesions along with the nasolabial cyst, nasopalatine duct cyst, and odontogenic keratocyst can be considered in the differential diagnosis of lesions occurring in the anterior maxilla.<sup>1</sup> The characteristic radiographic difference between dentigerous cyst and follicular AOT is that the radiolucency in the former is never associated with part of the root (always attached at the cervix) whereas in the latter it is most commonly associated with the part of the root.<sup>4</sup> AOT is usually surrounded by a well developed connective tissue capsule. It may present as a solid mass, a single large cystic space or as numerous small cystic spaces<sup>3</sup>.

The origin of the follicular variant can occur before or after cystic expansion (Cystic expansion in the jaw bone refers to the nature of expansion of the cyst through the buccal and lingual/palatal cortical plates). If it occurred after cystic expansion, then it effectively meant that the origin was from a dentigerous cyst and several such cases have been reported. If it occurred before cystic expansion, then the tumor tissue would fill the follicular space and the AOT would present as a solid tumor.<sup>1</sup>

The histology of AOT shows odontogenic epithelium with duct-like structures. Various sized of solid nodules of columnar or cuboidal epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue. The tumor may contain pools of amyloid like material and masses of calcification.<sup>1</sup> While AOT is reported as a tumor in the histological sign out, the notion that it represents a hamartomatous malformation adds a new dimension to its assorted histological architecture. Vimi S Mutalik reported as many as 20 different histological patterns of AOT in her article and had also noted few of them in her own case.<sup>2</sup> Although the tumor is odontogenic in origin, the reason for the occurrence of the ductal architecture is still hypothetical. Few authors believe it to be due to a cystic change in the follicles of tumor islands or probably an attempt to form glandular tissue since the origin is from the basal cells of the oral epithelium that has multiple differentiation capacity.<sup>2</sup> Of significant interest would be the occurrence of calcifying epithelial odontogenic tumor (CEOT) like area in AOT which was believed to be an altered phenotype in certain parts of the tumor. This hypothesis was further supported by an

immunohistochemical analysis using a panel of cytokeratin markers and vimentin, wherein the CEOT-like areas showed negative expression to CK 19.<sup>2</sup>

Marx and Stern considered AOT as a cyst and not a tumor and further gave a new terminology for this lesion, "Adenomatoid Odontogenic Cyst (AOC)." According to them, the AOC does not arise from the follicle of the tooth crown but instead arises from HERS, which would explain the finding of the tooth being completely within the lumen rather than the tooth root being within a bony crypt. Philipsen et al. have strongly argued in favour of the concept of AOT being derived from the complex system of dental laminae or its remnants.<sup>4</sup>

#### CONCLUSION –

Histopathology plays the most crucial role in diagnosing AOT. Its distinctive histologic features helps in arriving to an accurate diagnosis with ease. Even though it shows huge histologic diversity, the presence of an intact capsule reinforces its benign nature.

#### REFERENCES-

- 1) Pavitra Baskaran , Satyaranjan Misra , M. Sathya Kumar , R Mithra. Adenomatoid Odontogenic Tumor – A Report of Two Cases with Histopathology Correlation. J Clin Imaging Sci.2011; 1: 64.
- 2) Mutalik VS, Shreshtha A, Mutalik SS, Radhakrishnan R. Adenomatoid odontogenic tumor: A unique report with histological diversity. J Oral Maxillofac Pathol. 2012;16(1):118-21.
- 3) Vasudevan K , Kumar S.V. , Vigneswari S. Adenomatoid odontogenic tumor , an uncommon tumor. Contemp Clin Dent.2012;3:245-7.
- 4) Sonal Grover , Ahmed Mujib Bangalore Rahim , Nithin Kavassery, Parakkat , Shekhar Kapoor , Kumud Mittal , Bhushan Sharma et al. Cystic Adenomatoid Odontogenic tumor. Case reports in Dentistry. 2015.
- 5) Michael Prakasam , Saba Tiwari , Mrinal Satpathy, Vanaja Reddy Banda. Adenomatoid Odontogenic Tumour. BMJ Case Rep 2013.
- 6) Pulivarthi Sushma , MB Sowbhagya , P Balaji , C Poornima. Adenomatoid Odontogenic Tumor : A Case report. IJSS Case Reports & Reviews 2015;1(12):36-39.
- 7) Risk Diaz Castillejos , Ana Maria Nieto Munguia , Guillermina Castillo Ham. Adenomatoid odontogenic tumor. Case report and literature review. Revista Odontologica Mexicana 2015;19(3): 183-187.
- 8) Souza RS , Silva JBP , Villalba H , Santos CC , Georgevich Neto R , Giovanni EM. Adenomatoid odontogenic tumor : Case Report. J Dent Oral Care Med 3(2).
- 9) Neville BW, Damm DD, Allen CM, et al. Oral and maxillofacial pathology. 2nd edn. Philadelphia: WB Saunders, 2002:621–3.
- 10) Unal T, Cetingul E, Gunbay T. Peripheral adenomatoid odontogenic tumor: birth of a term. J Clin Pediatr Dent 1995;19:139–42.
- 11) Garg D, Palaskar S, Shetty VP, Bhushan A. Adenomatoid odontogenic tumor-hamartoma or True neoplasms: A case report. J Oral Sci. 2009;51:155–9.