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RESEARCH ARTICLE

IMPACTED MANDIBULAR DECIDUOUS AND PERMANENT CANINES: A 'DOUBLE TROUBLE' IN THE PATHOGENESIS OF ADENOMATOID ODONTOGENIC TUMOR: A CASE REPORT AND REVIEW OF LITERATURE.

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Abstract

Adenomatoid odontogenic tumor (AOT) is a benign non-invasive odontogenic lesion with slow growth potential and exceptionally low recurrence rate. This case report highlights an unusual case of an aggressive adenomatoid odontogenic tumor associated with impacted deciduous and permanent canines presenting in a 14 year old male in the anterior mandible.

Introduction:-

Odontogenic tumors represent a spectrum of lesions ranging from malignant (rare) and benign neoplasms to dental hamartomas, all arising from odontogenic residues, i.e. odontogenic epithelia and/or ectomesenchyme with variable amounts of dental hard tissues formed generally in the same sequence as in normal tooth development.[1]

The WHO histological typing of odontogenic tumors, jaw cyst and allied lesions (2005) has defined Adenomatoid odontogenic tumor (AOT) as "a tumor composed of odontogenic epithelium in a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth". [2]

AOT was first termed as '*epithelioma adamantinum'* by Steensland (1905), followed by Dreibladt (1907) as a '*pseudo adenoameloblastoma'*. It was later redesignated the term *'Adenomatoid odontogenic tumor'* by Philipsen and Birn (1969) and represents 3–7% of all odontogenic tumors. [3-5]

This tumor has sometimes been referred to as tumor of "two-third," because two-third of them occur in young females, two-third in the maxilla, two-third are associated with unerupted teeth, two-third are associated with canines and two-third of the tumors are diagnosed in the second decade of life.[5,6]

It has three well recognized clinicopathological variants- intrafollicular, extrafollicular and peripheral with 20 recognized histopathological variations. [4, 6, 7]

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Case report:-
A 14 year old male reported to the Goa Dental College and Hospital for evaluation of a gradually increasing swelling in relation to right lower jaw since two months.

Extra oral examination revealed a single, diffuse firm and non-tender swelling in relation to the right parasymphysis region measuring approximately 4 x 3 cms. Intra oral examination revealed a diffuse swelling in relation to 42, 44, 45, and 46 obliterating the buccal vestibule with expansion of lingual and buccal cortical plates. The swelling was hard, non- tender, non- pulsatile and non- fluctuant. The teeth in the area, 44, 45, and 46 tested non-vital. Orthopantomograph revealed a unilocular radiolucency crossing the midline and extending from 32 to 47, oval in shape with well-defined borders. Impacted deciduous and permanent canines were seen within the radiolucent lesion with radio-opaque flecks and resorption of the roots of overlying teeth. **[Figure 1]**

A provisional diagnosis of Adenomatoid odontogenic tumor was given.

Surgical excision of mass was followed by histopathological evaluation. On gross examination, the cut surface revealed granular hard tissue in relation to the crown of 43.

H/E stained sections showed odontogenic epithelium arranged in varied histoarchitectural patterns of ductules, rosettes, cell nests, sworls and sheets with a very scant connective tissue stroma surrounded by a fibrous capsule. Ductules and rosettes were lined by a single layer and double layer of cuboidal to low columnar cells respectively with vesicular hyperchromatic nuclei polarized towards the basement membrane. Centre of the ductules were clear, other areas showed eosinophilic coagulum. Nests and sheets of spindle to cuboidal cells were seen. Dystrophic calcifications in the form of spherical, ovoid and linear hematoxyphilic masses, some as Liesegang rings were seen. **[Figure 2]**

Research supporting hamartomatous nature	**Research supporting neoplastic nature**
Similar immunohistochemical composition of AOT with respect to reduced enamel epithelium and enamel organ (Amelogenin, enamelin, sheathelin, enamelysin) Ameloblastoma has greater proliferative potential than AOT Low Ki-67 expression No p16 expression More invasive behaviour of ameloblastomas compared to AOT Higher percentage of Ki-67 and Bcl-2 in solid ameloblastoma compared to AOT	Most odontogenic tumors are monoclonal Marked expression of cyclin D1 in AOT Spindle-shaped tumor cells of AOT show close associations with extracellular matrix signaling as well as cell proliferation Strong immunolocalization of HGF (hepatocyte growth factor and c-met in squamous cells present in AOT's Strong cytoplasmic expression of β-catenin Similar proliferative potential between ameloblastoma and AOT

Table I:-Literature relevant to biological behaviour of AOT

Features	**Typical findings of AOT**	**Unusual features present in our case**
Sex	Females	Male
Site	Anterior maxilla	Anterior mandible
Size	Does not exceed 1-3cm	4 x 3 cms
Root resorption	Extremely rare	Seen
Cortical plate penetration	Rare	Seen (lingual)
Impacted tooth	Usually one	Deciduous and permanent mandibular canines

Table II:-Unusual findings seen in our case related to typical features of an AOT

Figure 1:-a) Swelling in relation to the right parasymphysis region. b) Swelling in relation to 42, 44, 45, and 46 obliterating the buccal vestibule. c) Impacted deciduous and permanent canines seen within the radiolucent lesion with radio-opaque flecks in relation to the crown of the permanent canine and resorption of the roots of overlying teeth.

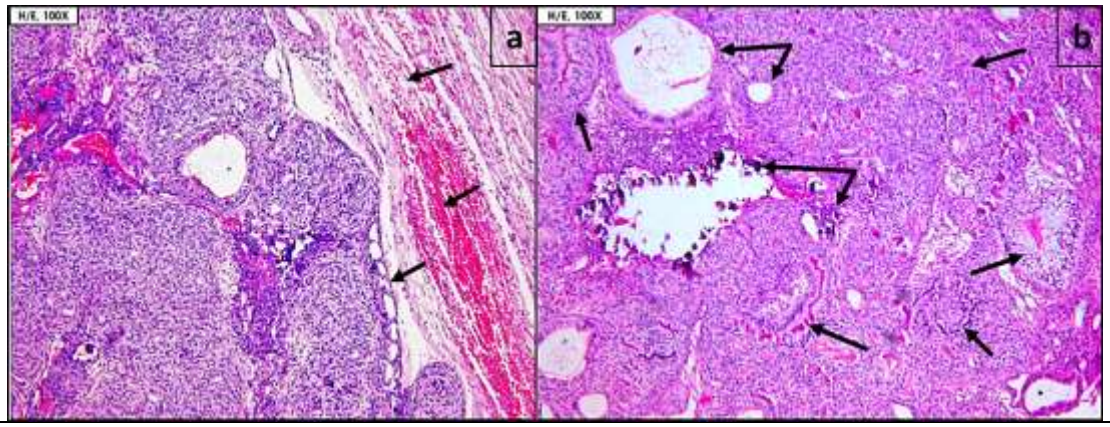

Figure 2:-a and b) Photomicrograph showing ductules, rosettes, cell nests, sworls and sheets of odontogenic epithelium with a very scant connective tissue stroma surrounded by a fibrous capsule.

Discussion:-

The Adenomatoid Odontogenic Tumor (AOT) is a benign (hamartomatous) non-invasive odontogenic lesion with slow growth potential and exceptionally low recurrence rate.

AOT's are said to arise from dental lamina remnants in the gubernacular cord of a developing permanent incisor, canine or premolar (96% cases) and from the epithelial lining of odontogenic cysts. [8, 9]

AOT's have 3 distinct clinical and radiographic variants: (a) Central (or intra-osseous) variant (b) Extra-follicular (or extra-coronal) variant and (c) Peripheral (or gingival) variant (PAOT): (i) 'peripheral AOT variant' (ii) 'erupted intra-osseous PAOT variant'. [4,6]

There is an ongoing debate whether AOT is a hamartoma or a true neoplasm. **[Table I]**

Immunohistochemically, the classical AOT phenotype is characterized by a cytokeratin (CK) profile similar to follicular cysts and/or oral or gingival epithelium based on positive staining with CK5, CK17 and CK19. [11, 12] Conservative surgical enucleation is the treatment modality of choice. [3]

Eighteen cases of mandibular AOT's in male patients have been reported in literature. Only one case with two impacted teeth has been reported in a female patient in anterior mandible. [6, 13-20] **[Table II]**

Recurrence of AOT is exceptionally rare and therefore the prognosis is excellent.[5] But in cases where unusual findings are seen, the follow up period should be extended to check for recurrences, as seen in the present case.

Conclusion:-

AOT is an uncommon odontogenic lesion, which can be readily identified from its histopathological features. Impacted deciduous teeth and unerupted succeeding permanent teeth, when associated with a swelling, always need to be investigated for odontogenic pathoses.

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