

Adenomatoid Odontogenic Tumour Presenting as Periapical Lesion: A Rare Case

Pavan Gujjar¹, Shylaja M.D², Nitin Patil³, Kailash Attur⁴

1- Senior Lecturer, Department Of Oral Pathology and Microbiology, Narsinhbhai Patel Dental College and Hospital, Visnagar, Gujarat. 2- Reader, Department Of Oral Pathology and Microbiology, Narsinhbhai Patel Dental College and Hospital, Visnagar, Gujarat. 3- Reader, Department of Oral and Maxillofacial Surgery, ACPM Dental College and hospital, Dhule, Maharashtra. 4- Reader, Department Conservative and Endodontics, Narsinhbhai Patel Dental College and Hospital, Visnagar, Gujarat.

Correspondence to:
Dr. Pavan Kumar, C-5/2, S.K Campus, Narsinhbhai Patel Dental College and Hospital, Ambaji Road, Visnagar, Gujarat. 384315
Contact us: www.ijohmr.com

ABSTRACT

The Adenomatoid odontogenic tumor (AOT) is a progressively growing asymptomatic benign non-invasive odontogenic tumor. A rare subvariant of the extrafollicular type of AOT may mimic a periapical lesion. We report a 28 year old male with a periapical lesion affecting maxillary right canine tooth. Surgical exploration of the lesion was done, and histopathological report was suggestive of AOT. This particular entity is very rare indeed, as literature showed only 9 such cases.

KEYWORDS: AOT, Extra follicular, Periapical

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a benign entity, which usually develops from enamel organ, dental lamina and reduced enamel epithelium or their remnants.¹ AOT is a benign (hamartomatous), non-invasive lesion with slow but progressive growth that accounts for 2.2–13% of all odontogenic tumors.

AOT effects most commonly in females during their second decade of life with the association of the anterior region of maxilla. There are three variants of AOT: follicular, extrafollicular and peripheral. The follicular type is a central intra bony lesion associated with an unerupted tooth, which accounts for about 70% of all cases. The extrafollicular variant is an intra-osseous lesion, but it is not related to any unerupted tooth, and usually represents around 25% of all AOT's. The rarest form of AOT is peripheral variant, which arises from gingival tissue. All three variants have the same histological aspect and clinical behaviour.² The follicular variant is associated with an impacted tooth (sometimes several teeth) and the maxillary canine is most often involved.³ The extrafollicular variant most commonly occurs in the maxilla either above the roots, in between the roots or superimposed on the roots of the teeth. The peripheral variant occurs almost exclusively in the anterior maxillary gingiva.⁴ Herewith we are presenting a case of extra follicular variant which was seen associated with a maxillary right canine.

CASE REPORT

A 28 year old male patient had reported with a complaint of swelling on the right side of the face since two months. The case was referred to the Department by a private practitioner, and hence a detailed history and clinical

picture and radiographs were not available. The only available history was that on extra oral examination obliteration of the nasolabial fold was noted. Vestibular obliteration and deep pocket of 6mm was seen in relation to the right maxillary canine. There was grade II mobility in relation to 13 and was non tender on vertical and horizontal percussion. The tooth was positive to vitality test. The provisional diagnosis of a globulomaxillary cyst, AOT, calcifying odontogenic cyst and OKC was considered because of its location.

Under local anesthesia, a bony window was created intraorally via a reflected mucoperiosteal flap. The lesion was removed easily from the adjoining bone with the involved tooth. The Surgical area was cleaned thoroughly and inspected for Oro-nasal or oro-antral communication which was not evident. The Flap was then sutured back with 3-0 silk suture.

The specimen was sent for histopathological examination. The gross specimen measured about 4x3 cm in size, grayish-black in colour and firm in consistency. The soft tissue was attached to the cervix of 13. The cut surface revealed a cystic lumen with a soft tissue growth filling most of the cystic space. (Fig. 1 a & b)

Multiple sections were taken, processed routinely, 4µ thickness was cut and stained with H&E. On histopathological examination, the tumor mass was encapsulated. Various-sized solid nodules of cuboidal or columnar epithelial cells formed ducts, nests and rosette-like structures. There was a presence of sheets of spindle cells between the rosette and tubular structures. Eosinophilic amorphous material and calcified masses were observed between spindles shaped cells and rosettes (Fig. 2 a & b). With the above histopathological findings

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a diagnosis of Adenomatoid odontogenic tumor (AOT) was made.



Fig- 1: a. The gross specimen was measuring about 4x3 cm in size, b. The cut surface revealed a cystic lumen with a soft tissue growth filling most of the cystic space.

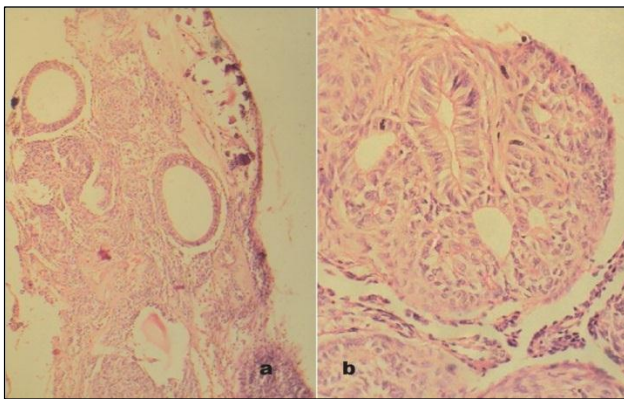


Fig-2: H & E stained section showing ducts and tumor islands a. 4 X magnification, b. 10 X magnification

DISCUSSION

In 1934 AOT was first described by Ghosh as an adamantinoma of the maxilla, and in 1948 Staphne was the first to recognize it as a distinct pathological entity. According to the second edition of the WHO 'Histological Typing of Odontogenic Tumors', AOT is defined as: "a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The tumor may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst."

Follicular and extrafollicular variants occur more commonly in the maxilla than in the mandible, with a ratio of 2.6:1. The female: male ratio for all age groups and AOT variants together is 2.3:1.⁵ Among the Asian populations higher female preponderance was seen (approximately 3:1).⁶ The AOT comprises approximately 3% of all odontogenic tumors, ranking behind odontoma, periapical cemental dysplasia (cementoma), myxoma and ameloblastoma.⁷

The origin of AOT is controversial,⁸ and evidence exist that this tumor could be derived from epithelial remnants of the dental lamina complex system. The lesion then presents radiographically as a residual, developmental,

lateral periodontal or radicular cyst depending on the location of epithelial cells of rests.

The extrafollicular variant is not associated with an erupted tooth like the follicular variant, and well defined, unilocular radiolucency is found between, above or superimposed on the roots of erupted teeth. It is characteristic that the rare sub variant mimicking a periapical lesion is, in fact, located palatally (or lingually) to the "involved" tooth.⁴ This case report illustrates characteristic clinical and radiographic features of the extra follicular variant of the AOT mimicking a periapical lesion in maxillary anterior region.

This particular sub variant is very rare indeed, as available literature reports only 9 cases.⁹ Out of these, 2 cases were presented in relation to mandibular canine, 1 case each in maxillary and mandibular premolar region, 2 cases in maxillary central incisor, 1 case in maxillary lateral incisor and 2 cases in mandibular molars. This is the only case where the lesion is present in Maxillary canine region. The maxillary incisor/canine region is often the site of an extrafollicular AOT in which the tumor produces a slow enlarging swelling.¹⁰ A distinct radiopaque border of the unilocular radiolucency is typical of the radiographic manifestation of an AOT.⁴

The lamina dura was not found to be intact around involved teeth an important finding that could suggest periapical radiolucency as periapical cyst or granuloma.¹¹ The AOT exhibits diverse histomorphologic features, but the light microscopic findings are remarkably consistent from tumor to tumor. Although present in varying proportions, the tumor represents a scattered duct like structures eosinophilic material, in several forms calcifications is seen and a fibrous capsule of variable thickness with a cellular multinodular proliferation of spindle cuboidal and columnar cells.¹²

Surgical enucleation with a conservative approach or curettage is a treatment of choice with a good prognosis.⁴

CONCLUSION

The extrafollicular variant of Adenomatoid odontogenic tumor presenting in the periapical region is a rare sub variant, only careful diagnostic procedures and adequate interpretation of clinical, radiographic and histopathological findings result in a correct diagnosis, which otherwise an incipient lesion will undergo an unnecessary endodontic treatment.

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