Case Report

Indian Journal of Dental Education
Volume 5 Number 4, October - December 2012

An Unusual Case of Adenomatoid Odontogenic Tumor Associated with Dentigerous Cyst

Sushruth Nayak, MDS*, Prachi Nayak, MDS, Rakesh Kumar Mannae, MDS***, Ashish Mahendra, MDS******

Abstract

Adenomatoid Odontogenic Tumor (AOT) is an uncommon, hamartomatous, benign, epithelial lesion of Odontogenic origin that was first described by Dreibaldt in 1970, as a pseudo adenoameloblastoma. The present World Health Organization (WHO) classification describes 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 occurs in both intraosseous and peripheral forms. Generally recurrence is very rare. The present case reports a 32 yr old male patient who presented with a chief complaint of swelling in the lower anterior region. On examination permanent lower right and left canine were missing with over retained lower right lateral incisor. Radiograph revealed a well defined unilocular radiolucent area in association with the lower anterior teeth. The lesion was excised along with the lower associated teeth and histopathological reports suggested AOT with the Dentigerous Cystic lining.

Key words: Adenomatoid odontogenic tumor; Dentigerous cyst.

Introduction

AOT is an epithelial tumor with an inductive effect on Odontogenic ectomesenchyme. It can be easily distinguishable from the classic intraosseous, infiltrative ameloblastoma. It was first described by Dreibaldt in 1907, as Pseudo-adenoameloblastoma and first reported by Harbitz in 1915 as a Cystic adamantinoma.[1] Philipsen and Brin[2] introduced the term Adenomatoid Odontogenic Tumor, which was accepted by WHO classification in 1971.[3] According to the second edition of WHO, 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".[4] The AOT appears in three types; 1) Follicular, 2) Extra follicular and 3) Peripheral. Follicular and Extra follicular type occurs within the bone or central variety which accounts for 97% of all AOT.[5] Adenomatoid Odontogenic Tumor is also called "two-thirds tumor", since 2/3rd of the tumor occurs in maxilla, 2/3rd occur in young females, 2/3rd of the tumor is associated with unerupted teeth and 2/3rd of the cases the affected teeth is canine.[6] AOT are benign and present a very low recurrence, making it unnecessary to carry out extensive and aggressive surgery. The surgical management of this tumor would be enucleation along with the associated impacted tooth.[5] Very few cases have been described that arise in association with dentigerous cyst. A systemic research of the English language medical literature revealed only seven such cases and only four cases of its occurrence in the

Author's Affilation: *Reader, Department of Oral and Maxillofacial Pathology & Microbiology, Chattisgarh Dental College and Research Institute, Rajnandgaon-491441, Chattisgarh.

**Senior lecturer, Department of Oral and Maxillofacial Pathology & Microbiology, Chattisgarh Dental College and Research Institute, Rajnandgaon-491441, Chattisgarh.

***Reader, Department of Oral Medicine and Radiology, Narayana Dental College, Nellore, Andhra Pradesh.

****Senior Lecturer, Department of Oral and Maxillofacial Pathology & Microbiology, Chattisgarh Dental College and Research Institute, Rajnandgaon-491441, Chattisgarh.

Reprints Requests: Dr. Sushruth Nayak, MDS, Reader, Department of Oral and Maxillofacial Pathology & Microbiology, Chattisgarh Dental College and Research Institute, Rajnandgaon-491441, Chattisgarh, INDIA. Phone: +919755443274

E-mail: drsushruthnayak@yahoo.co.in

Figure 1: Intra oral view

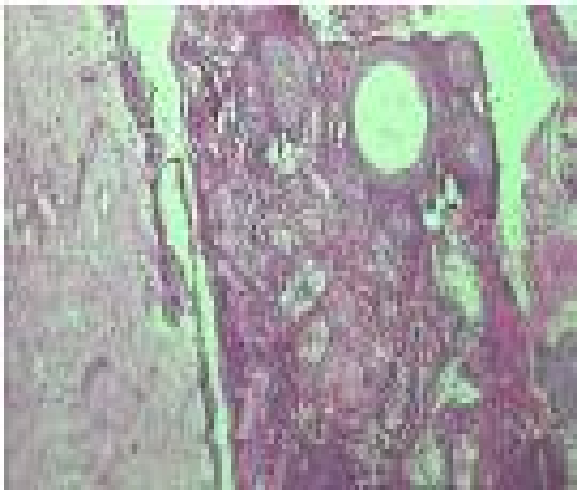
maxillary sinus. We present a case of AOT associated with dentigerous cyst in the maxillary canine region.

Case report

A 32 yr old male patient came with the chief complaint of swelling in the lower anterior region. Intra oral examination revealed swelling which was measuring 3x3 cm, spherical in shape and extending from lower primary left canine to lower primary right canine region. The permanent right and left canine were missing along with the presence of over retained primary lateral incisor (Figure 1). Radiograph revealed a well defined radiolucent area in association with lower anterior teeth. The permanent right and left canine were impacted (Figure 2). The tumor

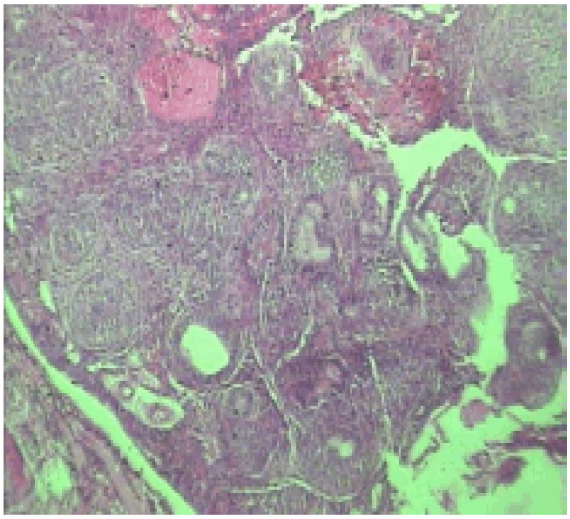
Figure 2: Orthopantamogram showing large radiolucent area

Figure 3: Thin cystic lining surrounding the lesion proper



was excised along with the associated and impacted tooth and the area was sutured. The specimen was put for histopathological examination. The H & E section revealed a well defined connective tissue capsule surrounding the lesion proper along with a thin cystic lining. The cystic lining was composed of 2-4 layers of epithelial cells resembling reduced enamel epithelium. The lesion proper showed duct like spaces lined by cuboidal cells. Other areas showed rosette like pattern and islands composed of spindle shaped cells. The connective tissue capsule revealed haphazardly arranged collagen fibers, few chronic inflammatory cells and blood vessels (Figure 3 & 4). The diagnosis of

Figure 4: Duct like structures and rosette pattern



Adenomatoid odontogenic tumor associated with dentigerous cyst was given.

Discussion

Adenomatoid odontogenic tumor is a slow growing lesion, constituting only 3% of all odontogenic tumors[7] and 0.1% of tumors of jaws[8] in general with a predilection for the anterior maxilla (ratio 2:1 relative to mandible) usually associated with impacted canine of young females in the second decade of life. In our case the lesion occurred in the anterior mandible associated with lower anterior teeth in a male patient in his third decade of life which is unusual. The origin of AOT is controversial. Some believe they originate from the odontogenic epithelium of a dentigerous cyst. Santos *et al*.[9,10] reported a case of AOT being developed in the fibrous capsule of the dentigerous cyst. Garcia Pola *et al*.[10,11] described the proliferation of an AOT in the epithelial border of a dentigerous cyst. Cassiano Francisco Weege *et al*.[9] reported a case of AOT associated with dentigerous cyst. The benign (hamartomatous) non-invasive AOT appears in 3 clinic topographic variants: 1) Follicular, 2) Extra follicular and 3) peripheral. Follicular and extra follicular variants are both intra-bony or central tumors and account for 97% of all AOTs of which 73% are of the follicular type.[5] The peripheral forms of the tumor are encountered very rarely. They usually appear as small sessile masses on the facial gingival of the maxilla. Clinically these lesions cannot be differentiated from the common gingival fibrous lesions.[12] In the present case AOT and dentigerous cyst are found in the same lesion. Very few cases have been described that arise in association with a dentigerous cyst. A systematic search of the English language medical literature revealed only seven such cases, and only four cases of its occurrence in the maxillary sinus. The hypothesis that follicular AOTs arise from the reduced enamel epithelium (REE) that lines the follicles of unerupted teeth is fairly conclusive and is supported by evidence that is both morphological and immuno-cytochemical in nature. They surround the crowns and are attached to the necks of unerupted teeth in a true follicular relationship. Many present as cystic lesions with only mural nodules of AOT lesional tissue and, in some instances, origin of the lesional tissue from the REE can be demonstrated histologically. Whether origin of the follicular variant occurs before or after cystic expansion has taken place is open to discussion. If it occurs after cystic expansion, then this effectively means origin from a dentigerous cyst, and several such case reports have been published.[10,13,14] If it occurs before cystic expansion, then the tumor tissue will fill the follicular space and the AOT will present as a solid tumor. Because of its low tendency to recur the tumor was enucleated surgically along with the associated and the impacted tooth.

References

1. Khot K, Pavitra AV. Mural adenomatoid odontogenic tumor in the mandible- A rare case. *Int J Oral Maxillofac Pathol.* 2011; 2: 35-9.
2. Philipsen HP, Birn H. The adenomatoid odontogenic tumor; Ameloblastic adenomatoid tumor or Adeno-Ameloblastoma. *Acta Pathol Microbial Scand.* 1969; 75; 375-98.
3. Pindborg JJ, Kramer IRH. WHO international histological classifications of tumors no.5 Histological typing of odontogenic tumors, jaw cysts and allied lesions.1st ed. Berlin; springer-verlag: 1971.
4. RH Kramer, JJ Pindborg, and M Shear. WHO International Histological Classification of Tumors. Histological Typing of Odontogenic Tumors. 2nd edition. Berlin Germany; Springer: 1992.
5. Philipsen HP, Reichart PA, Zhang KH, Nikai H, Yu Qx. Adenomatoid odontogenic tumor; biological profile based on 499 cases. *J Oral Pathol Med.* 1991; 20: 149-58.
6. 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.

7. Lee JK, Lee KB, Hwang BN. Adenomatoid odontogenic tumor: A case report. *J Oral Maxillofac Surg*. 2000; 58: 1161–4.
8. Ertunc D, Gelengul G, Murat B, Akif C. Adenomatoid odontogenic tumor: Case report and review. *Aust Dent J*. 1997; 42: 315–8.
9. Cassiano Francisco Weege Nonaka, Lelia Batista de Souza, Leda Bezerra Quindere. Adenomatoid odontogenic tumor associated with dentigerous cyst - unusual case report. *Braz J Otorhinolaryngol*. 2007; 73: 135–7.
10. Garcia-Pola Vallejo M, Gonzalez Garcia M, Lopez-Arranz JS, Herrero Zapatero A. Adenomatoid odontogenic tumor arising in a dental cyst: Report of an unusual case. *J Clin Pediatr Dent*. 1998; 23: 55–8.
11. Philipsen HP, Samman N, Ormiston IW, Wu PC, Reichart PA. Variants of adenomatoid odontogenic tumor with a note on tumor origin. *J Oral Pathol Med*. 1992; 21: 348–52.
12. Neville BW, Damm DD, Allen CM, Bouquot JE. Odontogenic cysts and Tumors. Oral and maxillofacial pathology. 3rd ed. Philadelphia: Elsevier; 2010, 713.
13. Tajima Y, Sakamoto E, Yamamoto Y. Odontogenic cyst giving rise to an adenomatoid odontogenic tumor: Report of a case with peculiar features. *J Oral Maxillofac Surg*. 1992; 50: 190–3.
14. Takahashi K, Yoshino T, Hashimoto S. Unusually large cystic adenomatoid odontogenic tumour of the maxilla: Case report. *Int J Oral Maxillofac Surg*. 2001; 30: 173–5.