Adenomatoid odontogenic tumor of maxilla in a 14-year-old child

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is an uncommon tumor of odontogenic origin, constituting only 0.1% of tumors and cysts of the jaw and 3% of all odontogenic tumors. A 14-year-old girl presented with right maxillary anterior swelling, facial asymmetry, continuously increase in size of swelling without any symptoms, and delayed eruption of permanent teeth. Orthopantomogram and computed tomography scan revealed a large unilocular radiolucency in right maxilla with permanent lateral incisor embedded within the lesion and permanent canine pushed away from its normal position. Cyst was enucleated completely under general anesthesia along impacted permanent teeth and retained deciduous teeth. Postoperative period was uneventful.

Key words: Adenomatoid odontogenic tumor, Hamartoma, Enucleation

Access this article online

Website: www.jpediatrdent.org

DOI: 10.4103/2321-6646.137692

Quick Response Code:

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is an uncommon tumor of odontogenic origin, first introduced by Steensland in 1905.[1] It was first described by Dreibladt, in 1907 as a pseudoameloblatoma.[2] In 1948, Stafne considered it as distinct pathological entity.[3] This lesion is known by many names, including adenoameloblastoma, ameloblastic adenomatoid tumor, adamantioma, epithelioma adamantium, or tertomatous odontoma.[4] Philipsen and Birn proposed the name adenomatoid odontogenic tumor in 1969, which was adapted by the WHO classification in 1971.[5] AOT constituting only 0.1% of tumors and cysts of the jaw and 3% of all odontogenic tumors.[6] Adenomatoid odontogenic tumor 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."[7] The incidence of AOT is higher in female[4] and usually located in the anterior region of maxilla within which and impacted tooth exists.[8] The tumor appear as an intra-oral-extra-oral swelling in the maxilla, and is sometimes referred as two-third tumor, because it occurs in the maxilla in about 2/3 cases, about 2/3 cases are in young females, 2/3 cases are associated with an unerupted tooth, and 2/3 affected teeth are canine. Although, it is generally believed that AOT is a hamartoma rather than a neoplasm, the lesion sometimes exhibits aggressive behavior such as becoming unusually large,[9] or spreading into the intracranial space.[10] Currently, it is generally accepted to be true neoplasm. Here we describe a case of AOT in 14-year-old female with details of aggressive clinical, radiographic, and histologic features suggesting that it was true neoplasm.

CASE REPORT

A 14-year-old girl presented with right maxillary anterior swelling of 2 months duration in dental Orthodontics and Pediatric Dentistry at University College of Medical Sciences and Guru Teg Bhadur Hospital Delhi. Patient had complained of facial asymmetry; continuously increase in size of swelling without any symptoms and delayed eruption of permanent teeth. Extra-oral examination disclosed large swelling on right anterior maxilla

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with obvious facial asymmetry. Intra-oral examination revealed a large compressible swelling in right anterior maxillary labial vestibule [Figure 1]. It was seen that deciduous maxillary right lateral incisor and canine were retained and their permanent successor teeth were missing. Orthopantomogram revealed a large unilocular radiolucency in right maxilla with permanent lateral incisor embedded within the lesion and permanent canine pushed away from its normal position [Figure 2]. Preoperative non contrast axial section of jaws and paranasal sinus revealed unilocular, expansile cystic lesion measuring 3 cm × 2.7 cm × 3 cm in right anteroinferior aspect of the maxilla. The lesion had shown punctuate "snow flake" calcification with unerupted permanent teeth lying within it. Cystic lesion seems to bulge into the nasal cavity causing superior displacement of inferior turbinate [Figure 3a]. Operative noncontrast enhanced computed tomography taken in coronal section demonstrate cystic nature of lesion and local expansive changes [Figure 3b]. A diagnostic aspiration was performed and 5 ml of blood mixed fluid withdrawn. Smear showed occasional squamous and histiocytes against hemorrhagic background. After correlating clinically and radiographically a provisional diagnosis of AOT was made.

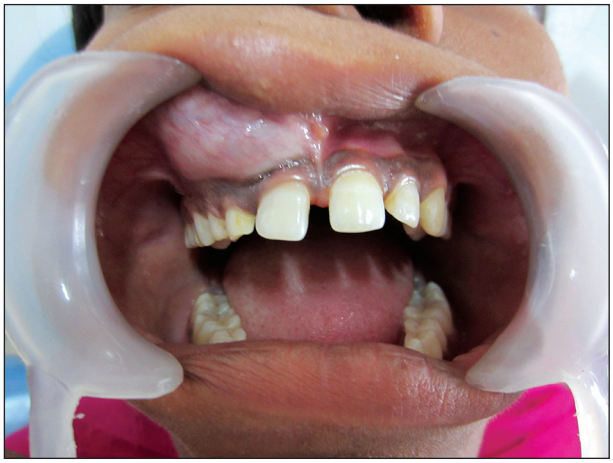

Figure 1: A large compressible swelling in right anterior maxillary labial vestibule

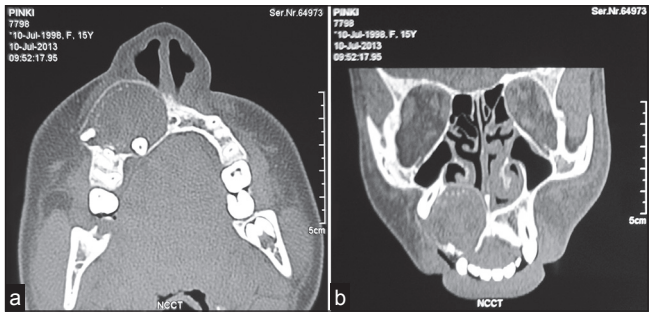

Figure 3: (a) Non-contrast computed tomography (CT) (axial cut) revealed unilocular, expansile cystic lesion in right anteroinferior aspect of the maxilla, (b) preoperative non-contrast enhanced computed tomography taken in coronal section demonstrate cystic nature of lesion and local expansive changes

A window has been created on the anterior surface of the cyst. The cyst lining is dissected off the surrounding bone, and the tooth is easily removed with the cyst.

The lesion was completely enucleated along with impacted lateral incisor and canine under general anesthesia. Retained deciduous lateral incisor and canine were also removed [Figure 4]. Postoperative course was uneventful. After surgery, specimen was sent for histopathological examination fixed in 10% formalin [Figure 5]. Histologic examination revealed the characteristics of AOT. The cyst was lined by odontogenic epithelium with occasional cellular projection in cystic lumen [Figure 6a]. Polyhedral and cuboidal epithelial cells in this stroma displayed characteristics duct-like structures filled with eosinophilic material [Figure 6b].

After 6 months of follow-up, a clinical and radiographic follow-up examination was performed. There was no evidence of recurrence and no apical resorption of adjacent teeth could be observed [Figure 7a and b].

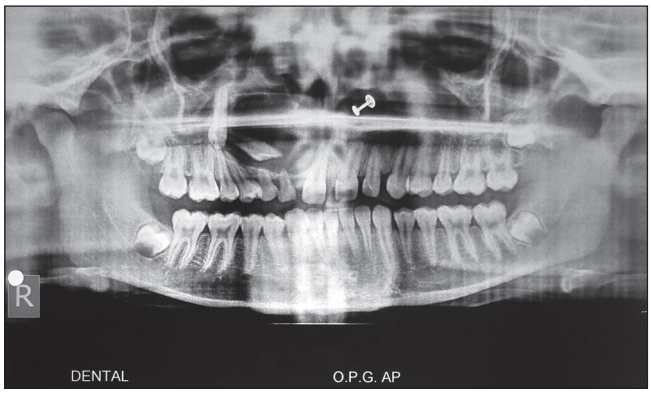

Figure 2: Orthopantamograph shows well-defined radiolucency tooth no. are 52 and 53 with 12 and 13 embedded in lesion and retained deciduous 54 and 53

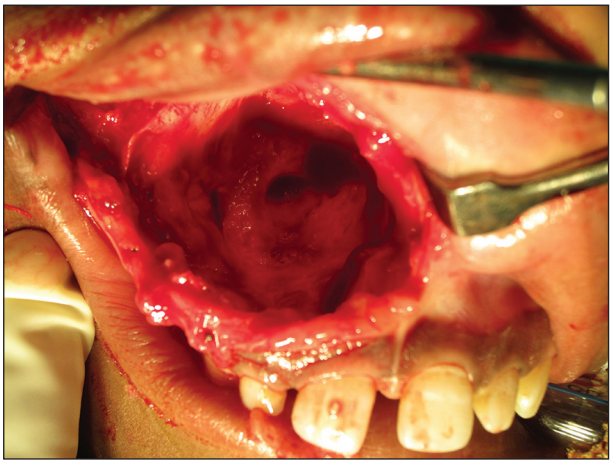

Figure 4: Complete enucleation of lesion along with removal of impacted lateral incisor and canine and retained deciduous lateral incisor and canine

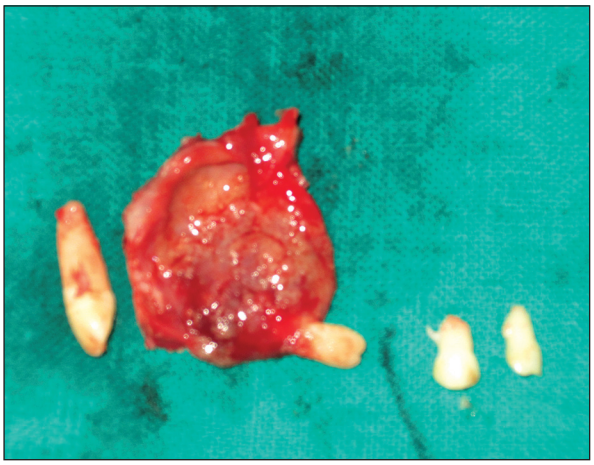

Figure 5: The enucleated adenomatoid odontogenic tumor with the associated maxillary 12 and 13 along with 52 and 53

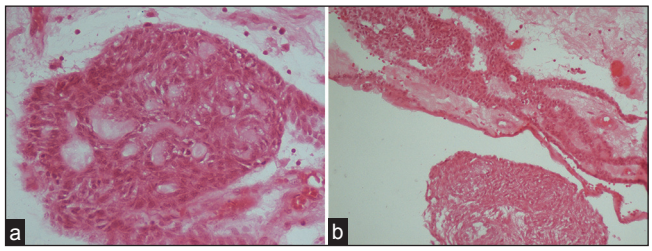

Figure 6: (a) Polyhedral and cuboidal epithelial cells in this stroma displayed characteristics duct-like structures filled with eosinophilic material. (H and E, stain ×400), (b) cystic wall lined by odontogenic epithelium with occasional cellular projection in cystic lumen (H and E, stain ×200)

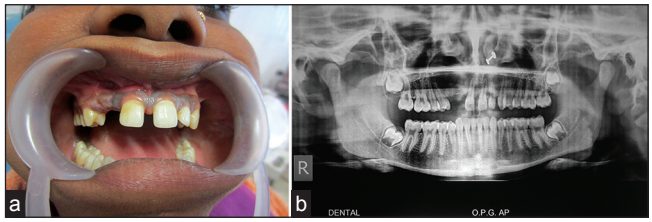

Figure 7: (a) Clinical follow-up after 6 months, (b) 6 months follow-up orthopantamograph

DISCUSSION

Steensland's in 1905 first reported AOT and termed as "epithelioma adamantinum." Since then a variety of terms have been used to describe this lesion of which the adeno-ameloblastoma was in common use for many years since the tumor was considered a histological variant of the ameloblastoma.[11] Philipsen and Birn in 1969 presented a review based on 76 cases of AOT, which showed the tumor to be an entity clearly distinguishable from the solid or multicystic ('classical') ameloblastoma.[5] They introduced the term AOT, to be adopted by the WHO in their "histological typing of odontogenic tumors, jaw cysts and allied lesions,"[12] and it is now the generally accepted nomenclature.

In the recent 2nd edition of the WHO "histological typing of odontogenic tumors" the AOT has been 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. It is generally believed that the lesion is not a neoplasm.[7]

Although larger lesions have been reported in the literature,[13,14] the tumors are usually in the dimensions of 1.5-3 cm.[15]

According to Philipsen and Reichart (1999) the AOT appears in three clinico-topographic variants: Follicular, extrafollicular and peripheral. The follicular and extrafollicular variants are both intrabony and account for approximately 96% of all AOTs of which 71% are of the follicular type.[11] The follicular type shows a well-defined, unilocular (round or ovoid) radiolucency associated with the crown and often part of the root of an unerupted tooth thus mimicking a dentigerous or follicular cyst.[5] The extrafollicular type is not associated with an unerupted tooth and the well-defined, unilocular radiolucency is found between, above or superimposed upon the roots of erupted, permanent teeth. These locations often lead to the preoperative, tentative diagnosis of a residual, radicular, "globulo-maxillary" or lateral periodontal cyst depending on the actual intraosseous site of the lesion.[11]

In approximately, two-thirds of the intrabony variants the radiolucency shows discrete foci having a flocculent pattern of scattered radiopacities. If the AOT contains minimal quantities of calcified deposits, intra-oral periapical radiographs are superior to panoramic radiographs in detecting the characteristic (although not pathogno-monic) radiopacities.[16]

The peripheral variant appears as a gingival fibroma or epulis attached to the labial gingiva. This type of AOT may show slight erosion of the alveolar bone crest but radiographic changes are often difficult to detect.[11]

Macroscopic

Macroscopically the intrabony AOT variants are roughly spherical in shape with a well-defined fibrous capsule. The cut surface may reveal a solid tumor mass or show one large or several small cystic spaces containing a yellowish, semi-solid material. In the follicular type a crown and often part of the root of an unerupted tooth is found embedded in the tumor mass or projecting into a cystic cavity.[17]

Microscopic

Irrespective of the tumor variants the histology is identical and exhibits a remarkable consistency. At low magnification

the most striking pattern is that of varying sized solid nodules of cuboidal or columnar epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue. Between the epithelial cells of nodules and in the center of the rosette-like configurations, eosinophilic amorphous material (often described as "tumor droplets") are present. Conspicuous within the cellular areas are structures of tubular or duct-like appearance. The duct-like spaces are lined by a single row of low columnar epithelial cells, the nuclei of which are polarized away from the lumenal surface. The lumen may be empty or contain a variable amount of eosinophilic material or cellular debris. The ducts vary considerably in diameter and may not always be present.[11]

Immunohistochemical

Tatemoto *et al.* demonstrated co-expression of keratin and vimentin in the tumor cells at the periphery of the ductal, tubular or whorled structures. Whereas tumor cells were positive for keratin stains, mineralized and hyaline material were negative.[18] Mori *et al.*[19] and Saku *et al.*[20] both studied enamel proteins in AOT and found amelogenin and enamelin in small mineralized foci, in the tumor cells and in the hyaline droplets.

Treatment and prognosis

Conservative surgical enucleation is the treatment modality of choice. For periodontal intrabony defects caused by AOT guided tissue regeneration with membrane technique is suggested after complete removal of the tumor.[21] Recurrence of AOT is exceptionally rare. Only three cases in Japanese patients are reported in which the recurrence of this tumor occurred.[22] Therefore, the prognosis is excellent.

REFERENCES

1. Steensland HS. Epithelioma adamantinum. J Exp Med 1905;6:377-89.
2. Lucas RB. Pathology of Tumors of the Oral Tissues. 4th ed. Edinburgh, Scotland: Churchill Livingstone; 1984. p. 66.
3. Stafne EC. Epithelial tumors associated with developmental cysts of the maxilla; a report of three cases. Oral Surg Oral Med Oral Pathol 1948;1:887-94.
4. Unal T, Cetingul E, Gunbay T. Peripheral adenomatoid odontogenic tumor: Birth of a term. J Clin Pediatr Dent 1995;19:139-42.
5. Philipsen HP, Birn H. The adenomatoid odontogenic tumour. Ameloblastic adenomatoid tumour or adeno-ameloblastoma. Acta Pathol Microbiol Scand 1969;75:375-98.
6. Khan MY, Kwee H, Schneider LC, Saber I. Adenomatoid odontogenic tumor resembling a globulomaxillary cyst: Light and electron microscopic studies. J Oral Surg 1977;35:739-42.
7. Kramer IR, Pindborg JJ, Shear M. WHO International Histological Classification of Tumors. Histological Typing of Odontogenic Tumors. 2nd ed. Berlin, Germany: Springer; 1992.
8. Neville BW, Damm DD, Allen CM, Bouquout JE. Oral and Maxillofacial Pathology. Philadelphia: Saunders; 1995. p. 529-30.
9. Mendis BR, MacDonald DG. Adenomatoid odontogenic tumour. A survey of 21 cases from Sri Lanka. Int J Oral Maxillofac Surg 1990;19:141-3.
10. Raubenheimer EJ, Seeliger JE, van Heerden WF, Dreyer AF. Adenomatoid odontogenic tumour: A report of two large lesions. Dentomaxillofac Radiol 1991;20:43-5.
11. Philipsen HP, Reichart PA. Adenomatoid odontogenic tumour: Facts and figures. Oral Oncol 1999;35:125-31.
12. Kramer IR, Pindborg JJ. WHO International Histological Classification of Tumors. No. 5. Histological Typing of Odontogenic Tumours, Jaw Cysts, and Allied Lesions. Berlin: Springer Verlag; 1971.
13. Larsson A, Swartz K, Heikinheimo K. A case of multiple AOT-like jawbone lesions in a young patient — A new odontogenic entity? J Oral Pathol Med 2003;32:55-62.
14. Khot K, Vibhakar PA. Mural adenomatoid odontogenic tumor in the mandible — A rare case. Int J Oral Maxillofac Pathol 2011;2:35-9.
15. Sandhu SV, Narang RS, Jawanda M, Rai S. Adenomatoid odontogenic tumor associated with dentigerous cyst of the maxillary antrum: A rare entity. J Oral Maxillofac Pathol 2010;14:24-8.
16. Dare A, Yamaguchi A, Yoshiki S, Okano T. Limitation of panoramic radiography in diagnosing adenomatoid odontogenic tumors. Oral Surg Oral Med Oral Pathol 1994;77:662-8.
17. Philipsen HP, Samman N, Ormiston IW, Wu PC, Reichart PA. Variants of the adenomatoid odontogenic tumor with a note on tumor origin. J Oral Pathol Med 1992;21:348-52.
18. Tatemoto Y, Tanaka T, Okada Y, Mori M. Adenomatoid odontogenic tumour: Co-expression of keratin and vimentin. Virchows Arch A Pathol Anat Histopathol 1988;413:341-7.
19. Mori M, Yamada K, Kasai T, Yamada T, Shimokawa H, Sasaki S. Immunohistochemical expression of amelogenins in odontogenic epithelial tumours and cysts. Virchows Arch A Pathol Anat Histopathol 1991;418:319-25.
20. Saku T, Okabe H, Shimokawa H. Immunohistochemical demonstration of enamel proteins in odontogenic tumors. J Oral Pathol Med 1992;21:113-9.
21. Berghagen N, Bergström J, Valerius-Olsson H. Adenomatoid odontogenic tumour. Periodontal aspects on diagnosis and treatment. Sven Tandlak Tidskr 1973;66:467-74.
22. Yazdi I, Nowparast B. Extraosseous adenomatoid odontogenic tumor with special reference to the probability of the basal-cell layer of oral epithelium as a potential source of origin. Report of a case. Oral Surg Oral Med Oral Pathol 1974;37:249-56.

How to cite this article: Kumar S, Khatri A, Kalra N, Tyagi R, Wadhwa N, Banga A. Adenomatoid odontogenic tumor of maxilla in a 14-year-old child. J Pediatr Dent 2014;2:61-4.

Source of Support: Nil. **Conflict of Interest:** None declared.