

CASE REPORT

Adenomatoid odontogenic tumor: A unique report with histological diversity

Vimi S Mutalik, Ashish Shreshtha, Sunil S Mutalik, Raghu Radhakrishnan

Department of Oral and Maxillofacial Pathology, Manipal College of Dental Sciences, Manipal University, Manipal, Karnataka, India

Address for correspondence:

Dr. Vimi S Mutalik,
Department of Oral Pathology, Manipal College
of Dental Sciences, Manipal University,
Manipal - 576 104, Karnataka, India.
E-mail: vimisunil@gmail.com

ABSTRACT

Odontogenic tumors are a group of heterogeneous lesions, features of which have been catalogued for several decades. Adenomatoid odontogenic tumor (AOT) is a relatively rare and distinct odontogenic tumor that is exclusively odontogenic epithelium in origin. Although considerable number of reports is available with regard to the clinical and histological spectrum of AOT, very few have highlighted its varied histological presentations. Therefore, this article focuses on the assorted histoarchitectural patterns of AOT.

Key words: Adenomatoid odontogenic tumor, dentigerous cyst, histomorphological patterns

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) rightfully called the master of disguise was first documented in literature by Steenslandas epitheliomaadamantinum.^[1] Over the years a variety of terminologies have been used to designate this extremely fascinating entity. Adenomatoid odontogenic tumor with its simple abbreviation AOT is the most widely accepted terminology Philipsen and Birn, 1969. The WHO histological typing of odontogenic tumors, jaw cyst and allied lesions (2005) has defined AOT as a tumor of odontogenic epithelium with duct-like structures and with varying degree of inductive changes in the connective tissue. The tumor may be cystic in its presentation or solid areas in some lesions may present as nodules in the capsule of a large cyst. 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. This unique report with a special emphasis on its histoarchitectural spectrum may augur our understanding of AOT biology.

CASE REPORT

A 14-year-old female presented to the clinic with a complaint of swelling on the left side of face accompanied with pain since 20 days. There was no history of trauma and extra

oral examination revealed mild facial asymmetry with the obliteration of the nasolabial fold. On palpation the swelling was bony hard, 2.0 × 2.5 cm in diameter with a well-defined border. Intraoral examination revealed obliteration of the mucobuccal fold with mild tenderness, while the oral mucosa was apparently unaffected. Hard tissue status revealed missing 23 and over retained 63. Submandibular bilateral lymphadenopathy was observed.

Radiographic examination showed a well-defined radiolucency with impacted 23 [Figures 1a and b]. A provisional diagnosis of dentigerous cyst with impacted 23 was made. Root resorption in relation to 63 was noted. The aspirated cystic fluid was straw colored and protein estimation level was 4.9 gm/dL. Grossly it was a globular mass, 2 × 2 × 2 cm, brownish black in color, firm in consistency with tooth attached to the specimen at the cervical area [Figure 1c].

Histopathology revealed cuboidal to columnar cells arranged in the form of nests and rosettes. Solid areas, duct-like pattern, whorled arrangement of cells, and tubular appearance is evident [Figure 2]. Convolved structures were noted and at the periphery of the lesion tumor cells are arranged in a strand-like configuration. Few cells were also arranged in a plexiform pattern and cribriform areas are also seen [Figure 3]. Lattice-work pattern is seen closer to the connective tissue capsule and foci of dense extravasated red blood cells were seen in few areas. Some amount of calcification, eosinophilic material, and leisegang ring formation was also observed [Figure 4].

A final diagnosis of adenomatoid odontogenic tumor was made. Treatment included enucleation under general anesthesia via crevicular incision with removal of the offending tooth. Healing was uneventful and the patient was lost to follow up.

Access this article online**Quick Response Code:**

Website:
www.jomfp.in

DOI:
10.4103/0973-029X.92987

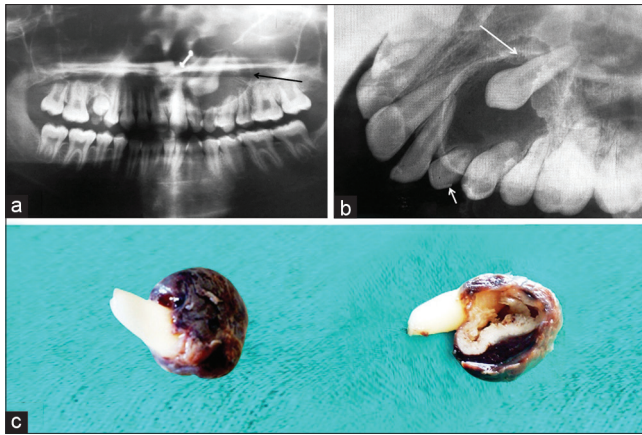


Figure 1: (a) Orthopantomograph shows well defined radiolucency with impacted 23. (b) Occlusal radiograph also shows well defined radiolucency with impacted 23. (c) Gross examination shows a globular mass, brownish black in color, firm in consistency with tooth attached to the specimen at the cervical area

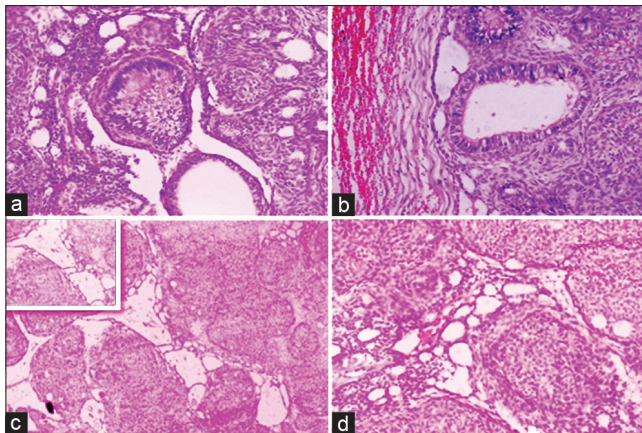


Figure 3: (a) Convoluted structure of tall columnar epithelial cells (H and E, ×10). (b) Strands of tumor cells 1 to 2 cell layer thick at the periphery (H and E, ×10). (c) Thin anastomosing strands of basoid cells in a plexiform pattern (H and E, ×10). Inset showing high power view of the same (H and E, ×20). (d) Cellular areas with cribriform configuration (H and E, ×10)

DISCUSSION

The case is of specific interest because of the vivid histological architecture it is presented with. However, an intensive literature search revealed very few reports where only the patterns have been showcased. Therefore, we have tried to revive various views of the authors with regard to the histomorphological spectrum.

AOT usually presents with classical clinical features; however, a multicentric study has shown an increased frequency of AOT in the Nigerian population which is as high as 39%.^[1] This case was seen in female in the second decade of life that coincides well with the statistical observation. There are reports of AOT presenting in infants^[2] and also in individuals in eighth decade of life.^[3] AOT involves both the bone and soft tissue in anatomic configuration.^[2,4] Our case followed the

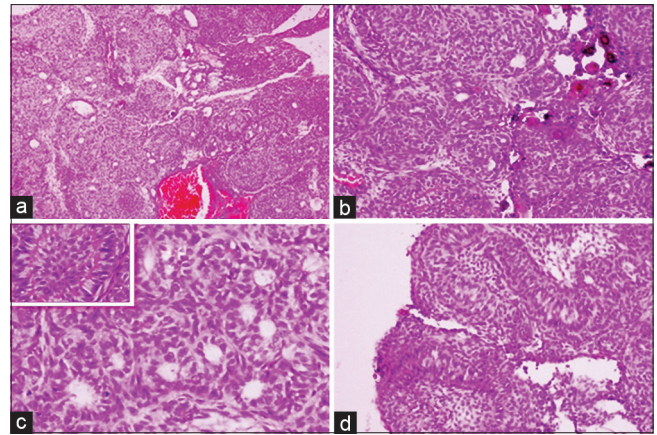


Figure 2: (a) Cellular multinodular proliferation of cells in the form of nests and rosettes. (H and E, ×4). (b) Solid areas of cells in the form of whorls interspersed with areas of calcification. (H and E, ×10). (c) Duct-like pattern lined by cuboidal cells (H and E, ×20), inset showing the characteristic rosette like structure (H and E, ×40). (d) Curved rows of cells giving a tubular appearance (H and E, ×10)

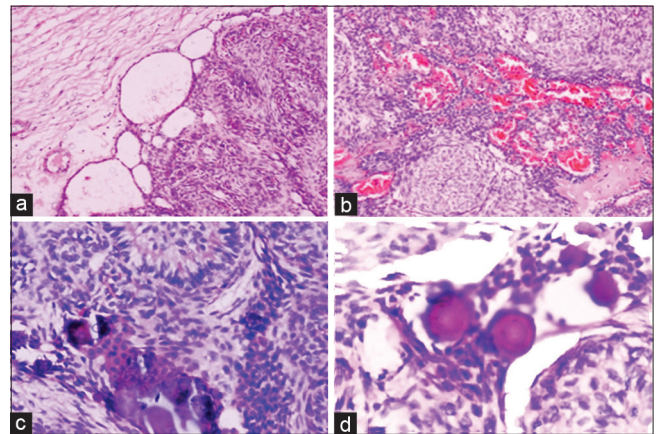


Figure 4: (a) Lattice work pattern seen closer to the connective tissue capsule (H and E, ×10). (b) Dense hemorrhagic areas (H and E, ×10). (c) Irregular calcified bodies amidst the tumor cells (H and E, ×10). (d) Leisegang pattern of calcification in the form of globular or spheroidal masses (H and E, ×10)

biological trend of the common intraosseous location in the maxillary canine region. Cases have also been reported in the mandible,^[5] molar areas,^[2,6] in maxillary sinus^[7] and along with embedded primary teeth.^[8]

Although AOT is called the perfect imitator of the dentigerous cyst, the characteristic clinical features renders the diagnosis relatively obvious. Reports have also shown the size of the lesion to be as large as 12 cm.^[3] It has a decisive sex predilection for females, while few cases have also been seen in males.^[2] The highest female to male ratio is seen in Japanese population, the reason for which is unknown.^[8] As far as the duration of the lesion is concerned there are cases which have been present since 37 years.^[4] Moreover, the presence of an intact capsule in most of the cases further reinforces the benign nature of AOT. This tumor is also associated with dilacerated tooth; anomalous tooth forms^[3] and supernumerary teeth.^[9] Garg *et al.*

have reported a case of unencapsulated AOT that caused root resorption and was fast growing in nature.^[10] But few others have reported AOT showing displacement of teeth,^[8] buccal plate perforation, hypesthesia,^[2] and root resorption.^[6] The gross features in our case was in accordance to the previously reported cases in literature but the striking feature was the attachment of this lesion exactly to the cemento-enamel junction that is quite unusual in most of the cases when examined grossly. Radiologically it has unilocular radiolucency but multilocular variant are also reported which gives credence to the occurrence of multiple AOT.^[3] Considerable amount of debate is still going on whether to consider AOT as a hamartoma or neoplasm. The relatively small size of the tumor and lack of recurrences in most cases support the fact that it is a hamartoma. On the contrary, few authors suggest that early detection could be the reason for small size of the lesion.^[10] Therefore, an increased variation as compared to odontogenic apparatus and aggressive features in few of the reported cases certainly gives credibility for the neoplastic origin.

As many as 20 different histological patterns of AOT have been described in the literature, yet there are not many reports that discuss its assorted presentation [Table 1].

The most interesting finding in our case was that majority of the above mentioned histomorphological patterns existed in one

Table 1: Histological patterns of AOT

Histological diversity	Reference with year
AOT with CEOT-like areas	(Philipsen <i>et al.</i> , ^[8] 1991)
Arrangement of cells in layers	(Garg <i>et al.</i> , ^[10] 2009)
Convoluting cords or bodies mimicking invaginations	(Philipsen <i>et al.</i> , ^[8] 1991)
Cribriform pattern	(Takahashi <i>et al.</i> , ^[11] Philipsen <i>et al.</i> , ^[8] 1991)
Cystic variant of AOT	(Gadeawarand Srikant, ^[12] 2010)
Cystic variants forming mural lining	(Siar <i>et al.</i> , ^[13] 1986)
Duct-like structures	(Philipsen <i>et al.</i> , ^[8] 1991) (Lee <i>et al.</i> , ^[9] 2000)
Interlacing strands of cells	(Philipsen <i>et al.</i> , ^[8] 1991)
Luminal proliferations into the cystic lumen	(Philipsen <i>et al.</i> , ^[8] 1991)
Nests-like pattern	(Philipsen <i>et al.</i> , ^[8] 1991)
Peripheral strand of smaller cells which form net like proliferations	(Fredrich <i>et al.</i> , ^[7] 2009)
Ribbon-like pattern	(Garg <i>et al.</i> , ^[10] 2009)
Ring-like pattern of tumor cells	(Garg <i>et al.</i> , ^[10] 2009)
Rosette-like arrangement	(Philipsen <i>et al.</i> , ^[8] 1991)
Sheets of tumor cells	(Philipsen <i>et al.</i> , ^[8] 1991)
Sieve-like pattern	(Takahashi <i>et al.</i> , ^[11])
Solid nodules of cells	(Takahashi <i>et al.</i> , ^[11])
Trabecular arrangement	(Philipsen <i>et al.</i> , ^[8] 1991)
Tubular arrangement	(Philipsen <i>et al.</i> , ^[8] 1991)
Whorled spheroidal masses of tumor cells	(Nomura <i>et al.</i> , ^[2] 1992) (Garg <i>et al.</i> , ^[10] 2009)

particular case. Calcification was seen in the form of irregular masses, leisegang rings, spheroidal, and globular forms. 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.^[14] Moreover, the occurrences of all these patterns could just be representing the caricatures of the enamel organ itself. 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.^[15] Few authors even suggested that these areas could just be considered the normal histoarchitectural spectrum of AOT.^[16]

Other than the above patterns histology also shows presence of capsule,^[4,13] necrosis, hyalinization,^[4] melanin pigmentation,^[6] and dysplastic dentinoid, osteodentin, calcification. The presence of mitotic figures,^[2] increased vascularity,^[2] nuclear pleomorphism, degeneration^[8] and nuclear hyperchromatism^[7] has also been reported. Calcifications exhibited in the form of leisegang rings,^[8] spheroidal masses,^[13] and globular forms.^[8] Treatment usually involves removal of the tumor in toto. Recurrence is very unusual.

CONCLUSION

Therefore, this article has presented tumor profile on varied histomorphological variants of AOT. However, what is intriguing is that irrespective of the pattern the biological behavior of the tumor never changed unlike that of the other tumors. Moreover, histology has always remained identical with remarkable consistency. Due to this distinctive histomorphology the diagnosis can always be made with ease. This addendum calls attention for stressing more on the histomorphology of AOT and in knowing why none of these patterns have any effect on the biological behavior of these tumors.

REFERENCES

- Mohamed A, Singh AS, Raubenheimer EJ, Bouckaert MM. Adenomatoid odontogenic tumour: Review of the literature and an analysis of 33 cases from South Africa. *Int J Oral Maxillofac Surg* 2010;39:843-6.
- Nomura M, Tanimoto K, Takata T, Shimosato T. Mandibular adenomatoid odontogenic tumor with unusual clinicopathologic features. *J Oral Maxillofac Surg* 1992;50:282-5.
- Tsaknis PJ, Carpenter WM, Shade NL. Odontogenic adenomatoid tumor: Report of case and review of the literature. *J Oral Surg* 1977;33:146-9.
- Bedrick AE, Solomon MP, Ferber I. The adenomatoid odontogenic tumor: An unusual clinical presentation. *Oral Surg*

- Oral Med Oral Pathol 1979;48:143-5.
5. Handschel JG, Depprich RA, Zimmermann AC, Braunstein S, Kübler NR. Adenomatoid odontogenic tumor of the mandible: Review of the literature and report of a rare case. *Head Face Med* 2005;24:1-3.
 6. 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.
 7. Friedrich RE, Scheuer HA, Zustin J. Adenomatoid odontogenic tumor (AOT) of maxillary sinus: Case report with respect to immunohistochemical findings. *In vivo* 2009;23:111-6.
 8. Philipsen HP, Reichart PA, Zhang KH, Nikai H, Yu QX. Adenomatoid odontogenic tumor: Biologic profile based on 499 cases. *J Oral Pathol Med* 1991;20:149-58.
 9. Lee JK, Lee KB, Hwang BN. Adenomatoid odontogenic tumor: A case report. *J Oral Maxillofac Surg* 2000;58:1161-4.
 10. Garg D, Palaskar S, Shetty VP, Bhushan A. Adenomatoid odontogenic tumor - hamartoma or true neoplasm: A case report. *J Oral Sci* 2009;51:155-9.
 11. Takahashi H, Fujita S, Shibata Y, Yamaguchi A. Adenomatoid odontogenic tumour: Immunohistochemical demonstration of transferrin, ferritin and alpha-one-antitrypsin. *J Oral Pathol Med* 2001;30:237-44.
 12. Gadewar DR, Srikant N. Adenomatoid odontogenic tumour: Tumour or a cyst, a histopathological support for the controversy. *Int J Pediatr Otorhinolaryngol* 2010;74:333-7.
 13. Siar CH, Ng KH. Adenomatoid odontogenic tumor: A survey of 44 new cases in Malaysia. *Ann Dent* 1986;45:11-4.
 14. Oehlers FA. The so called adenoameloblastoma. *Oral Surgery Oral Med Oral Pathol* 1961;14:712-25.
 15. Leon JE, Mata GM, Fregnani ER, Carlos-Bregni R, de Almeida OP, Mosqueda-Taylor A, et al. Clinicopathological and immunohistochemical study of 39 cases of Adenomatoid Odontogenic Tumor: A multicentric study. *Oral Oncol* 2005;41:835-42.
 16. Montes Ledesma C, Mosqueda Taylor A, Romero de León E, de la Piedra Garza M, Goldberg Jaukin P, Portilla Robertson J. Adenomatoid odontogenic tumour with features of calcifying epithelial odontogenic tumour. (The so-called combined epithelial odontogenic tumour.) Clinico-pathological report of 12 cases. *Eur J Cancer B Oral Oncol* 1993;29B:221-4.

How to cite this article: Mutalik VS, Shreshtha A, Mutalik SS, Radhakrishnan R. Adenomatoid odontogenic tumor: A unique report with histological diversity. *J Oral Maxillofac Pathol* 2012;16:118-21.

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