

Adenomatoid Odontogenic Tumor: A Dentigerous Cyst Mimic in Maxilla

A L Hemalatha¹, S N Shobha², C R Raghuveer³, Swati Sahni³, Amita Kumari³

¹Professor and Head, Department of Pathology, Adichunchanagiri Institute of Medical Sciences, Mandya, Karnataka, India, ²Assistant Professor, Department of Pathology, Adichunchanagiri Institute of Medical Sciences, Mandya, Karnataka, India, ³Post-graduate, Department of Pathology, Adichunchanagiri Institute of Medical Sciences, Mandya, Karnataka, India

Abstract

The adenomatoid odontogenic tumor is a rare and distinct neoplasm which is considered a slowly progressive, benign, non-neoplastic hamartomatous lesion by many. It is notoriously misdiagnosed as a dentigerous cyst. It accounts for 1-9% of odontogenic tumors. The most common site of occurrence is maxilla and it is usually associated with an unerupted, permanent tooth. It is most commonly seen in young women patients. It closely resembles ameloblastoma or dentigerous cyst. A rare case of adenomatoid tumor of maxilla mimicking a dentigerous cyst is presented here.

Key words: Adenomatoid odontogenic tumor, Dentigerous cyst, Odontogenic cyst

INTRODUCTION

The adenomatoid odontogenic tumor is a relatively uncommon tumor of odontogenic epithelium comprised of duct-like structures. It may be partly cystic and partly solid in consistency. It has been called by various names after being described for the 1st time by Steensland.¹

Drebaladt called it pseudo adenoameloblastoma,² Harbitz "cystic adamantinoma"³ and Ghosh "adamantinoma."⁴ It was recognized as a distinctive entity by Stafne.⁵ It was eventually recognized that it is not a type of ameloblastoma.⁶ Gorlin called it ameloblastic adenomatoid tumor while Abrams *et al.*, suggested the name ameloblastic adenomatoid tumor⁷ and adopted this term in the WHO histological typing of tumor.

Clinical Summary

A 17-year-old male patient presented with progressive swelling on the left side of the face which was present since 6 months.

Local examination revealed a diffuse swelling involving the left infraorbital and malar areas extending up to the left nasolabial fold, mid-zygomatic arch, and infraorbital margin.

Computed tomography scan findings showed an expansile radiolucent lesion with a well-defined radiosclerotic margin enwrapping a tooth within it.

A provisional clinical diagnosis of the dentigerous cyst was offered.

Enucleation was done and the specimen submitted for histopathological examination.

Gross Examination Findings

Single, globular tissue mass measuring 4 cm × 3 cm × 1.5 cm. External surface was gray brown. A tooth was embedded in the soft tissue mass (Figure 1). The cut surface was gray-white to gray-brown with firm solid areas (Figure 2).

Microscopy

A benign epithelial tumor with a round to oval tumor cells arranged in nodules, whorls, and ductular patterns was seen. The tumor cells showed scant cytoplasm and uniform nuclei with bland chromatin. The epithelial nodules were surrounded by polyhedral to spindle-shaped cells (Figure 3). Intervening areas of homogenous hyaline material and calcified deposits were seen (Figure 4).

Access this article online



www.ijss-sn.com

Month of Submission : 11-2015
Month of Peer Review : 12-2015
Month of Acceptance : 01-2016
Month of Publishing : 01-2016

Corresponding Author: Dr. A L Hemalatha, G-3 Sai Brindavan Apartments, 63/64 Industrial Suburb Vishwesharanagar, Mysore - 570 008, Karnataka, India. Phone: +91-8453399335. E-mail: halingappa@gmail.com

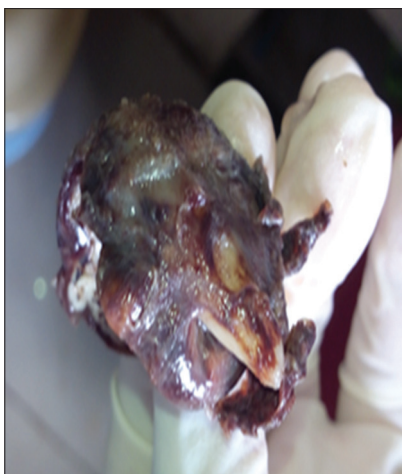


Figure 1: Globular, soft tissue mass with an embedded tooth

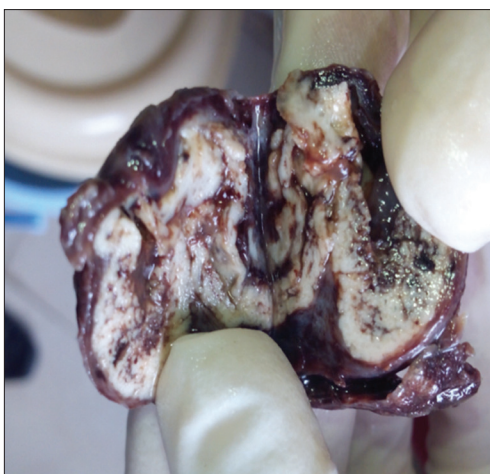


Figure 2: Cut surface - encapsulated gray - white to grey - brown tumor with firm solid areas

Final Diagnosis

Adenomatoid Odontogenic Tumor.

DISCUSSION

The adenomatoid odontogenic tumor is an uncommon neoplasm.⁸ Three variants of the tumor have been described namely, intraosseous follicular, intraosseous extrafollicular and peripheral types. The extrafollicular type shows no relation with an impacted tooth which is seen in about 24% of cases as in the present case.⁹ The majority of the tumors (73%) are of a follicular type and are seen in association with an unerupted tooth. The peripheral variant (3%) is seen attached to the gingival structures. The intraosseous follicular and extrafollicular types are commoner in the maxilla than in the mandible.^{10,11} The tumor is more common seen in females with a female to male ratio of 2:1.¹² The histogenesis of this tumor is debatable.^{13,14} It is suspected

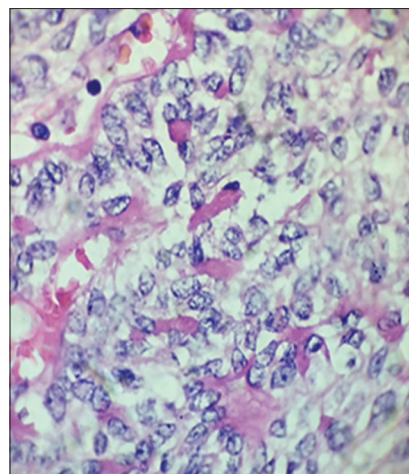


Figure 3: Epithelial nodules surrounded by polyhedral to spindle shaped cells (H and E $\times 45$)

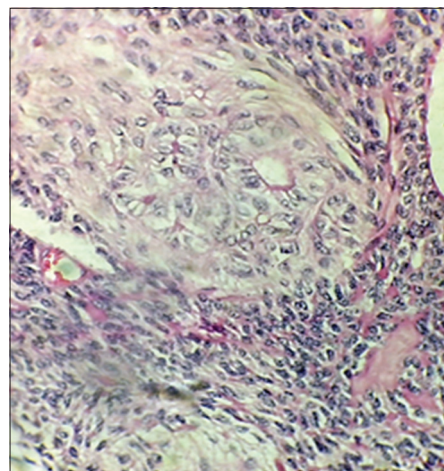


Figure 4: Tumor cells with scant cytoplasm, uniform nuclei, and bland chromatin. Intervening areas of homogenous hyaline material and calcified deposits

to arise from odontogenic epithelium because it arises more commonly in the teeth bearing bones.¹⁵ The tumors are usually small ranging from 1.5 to 3 cm.¹⁶ Larger tumors as in the present case have also been reported.^{7,17} Sometimes extensive calcification can be seen in the tumor.⁷ The clinico-radiologic appearances are often mistaken for calcifying odontogenic tumor or dentigerous cyst as in the present case. The differential diagnoses include ameloblastoma, ameloblastic fibro-odontoma, and ameloblastic fibroma.¹⁸

Since the tumor is well-encapsulated and is known to have a benign behavior, conservative surgical enucleation is the preferred treatment of choice. It has an excellent outcome without any recurrence.¹⁸ Our patient who underwent surgical enucleation 6 months ago is asymptomatic on follow-up.

CONCLUSION

The adenomatoid odontogenic tumor is an uncommon entity which is often clinically mistaken for the dentigerous or odontogenic cyst. Histopathology is the gold standard in its diagnosis. Conservative surgical treatment is curative.

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, UK: Churchill Livingstone; 1984.
3. Harbitz F. On cystic tumours of the maxilla, and especially on adamantine cystadenomas (Adamantomas). *Dent Cosm* 1915;57:1081-93.
4. Ghosh LS. Adamantinoma of the upper jaw: Report of a Case. *Am J Pathol* 1934;10:773-90.5.
5. 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.
6. Rick GM. Adenomatoid odontogenic tumor. *Oral Maxillofac Surg Clin North Am* 2004;16:333-54.
7. Nigam S, Gupta SK, Chaturvedi KU. Adenomatoid odontogenic tumor – A rare cause of jaw swelling. *Braz Dent J* 2005;16:251-3.
8. Philipsen HP, Srisuwan T, Reichart PA. Adenomatoid odontogenic tumor mimicking a periapical (radicular) cyst: A case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002;94:246-8.
9. Philipsen HP, Reichart PA. Adenomatoid odontogenic tumour: Facts and figures. *Oral Oncol* 1999;35:125-31.
10. Yilmaz N, Acikgoz A, Celebi N, Zengin AZ, Gunhan O. Extrafollicular adenomatoid odontogenic tumor of the mandible: Report of a case. *Eur J Dent* 2009;3:71-4.
11. Ajagbe HA, Daramola JO, Junaid TA, Ajagbe AO. Adenomatoid odontogenic tumor in a black African population: Report of thirteen cases. *J Oral Maxillofac Surg* 1985;43:683-7.
12. Giansanti JS, Someren A, Waldron CA. Odontogenic adenomatoid tumor (adenoameloblastoma). Survey of 111 cases. *Oral Surg Oral Med Oral Pathol* 1970;30:69-88.
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. Bravo M, White D, Miles L, Cotton R. Adenomatoid odontogenic tumor mimicking a dentigerous cyst. *Int J Pediatr Otorhinolaryngol* 2005;69:1685-8.
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. 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.
17. Khot K, Vibhakar PA. Mural adenomatoid odontogenic tumor in the mandible - A rare case. *Int J Oral Maxillofac Pathol* 2011;2:35-9.
18. Motamedi MH, Shafeie HA, Azizi T. Salvage of an impacted canine associated with an adenomatoid odontogenic tumour: A case report. *Br Dent J* 2005;199:89-90.

How to cite this article: Hemalatha AL, Shobha SN, Raghuveer CR, Sahni S, Kumari A. Adenomatoid Odontogenic Tumor: A Dentigerous Cyst Mimic in Maxilla. *Int J Sci Stud* 2016;3(10):174 –176.

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