

Adenomatoid Odontogenic Tumour: Review and Case Report

(Tumeur odontogène adénomatoïde : examen et rapport de cas)

- Puneet Batra, BDS, MDS, MOrth RCS(Edinburgh), DNB, PGDHM, PGDMLS •
- Shyam Prasad, BDS, MDS, PGDMLS •
- Hari Parkash, MDS •

S o m m a i r e

La tumeur odontogène adénomatoïde est une lésion bénigne hamartomateuse non invasive dont la croissance progressive est lente. Ses 3 variantes – folliculaire, extrafolliculaire et périphérique – présentent les mêmes caractéristiques histologiques. Le présent rapport décrit une patiente ayant une importante tumeur odontogène adénomatoïde de la mandibule, où la canine mandibulaire atteinte est poussée contre le côté controlatéral. L'article donne aussi des renseignements actualisés aux dentistes généralistes au sujet de divers aspects du diagnostic de la tumeur, met en évidence les controverses à propos de son origine et traite de sa prise en charge à la lumière de découvertes récentes.

Mots clés MeSH : case report; mandibular neoplasms/pathology; odontogenic tumors/pathology

© J Can Dent Assoc 2005; 71(4):250-3
Cet article a été révisé par des pairs.

Adenomatoid odontogenic tumour was first described by Dreibladt, in 1907, as a pseudo-ameloblastoma.¹ In 1948 Stafne² considered it a distinct entity, but it was classified by others as a variant of ameloblastoma. As a result, the lesion is known by many names, including adenoameloblastoma, adenoameloblastic odontoma, epithelial tumour associated with developmental cysts, ameloblastic adenomatoid tumour and adenomatoid or pseudoadenomatous ameloblastoma.²⁻⁴ Philipsen and Birn⁴ proposed the name adenomatoid odontogenic tumour in 1969 and suggested that it not be regarded as a variant of ameloblastoma because of its different behaviour. This term was adopted by the World Health Organization (WHO) classification⁵ in 1971.

There are 3 variants of adenomatoid odontogenic tumour,⁶⁻⁸ the follicular type (accounting for 73% of cases), which has a central lesion associated with an embedded tooth; the extrafollicular type (24% of case), which has a central lesion and no connection with the tooth; and the peripheral variety (3% of cases). Both types of central intraosseous tumours produce a corticate radiolucency, sometimes with radiopaque specks. The follicular type is usually initially

diagnosed as a dentigerous or follicular cyst. The extrafollicular type usually presents as a unilocular, well-defined radiolucency found between, above or superimposed on the roots of erupted teeth and often resembling a residual, radicular, globulomaxillary or lateral periodontal cyst. The peripheral type usually presents as a gingival swelling, located palatally or lingually relative to the involved tooth. In two-thirds of cases of the types with a central lesion the radiolucency shows discrete foci and a flocculent pattern of scattered radiopacities. If the tumour has minimum quantities of calcified deposits, intraoral periapical radiography is superior to panoramic radiography in detecting the characteristic radiopacities (although these are not pathognomic).⁹

This report describes an unusually large follicular adenomatoid odontogenic tumour in the mandible, illustrates the clinical, microscopic and biological features of the tumour and emphasizes the importance of the relation between the dental follicle and the tumour tissue. The article provides a refresher for general dental practitioners about diagnostic aspects of this tumour and discusses current controversies in pathogenesis and management.



Figure 1: Intraoral photograph shows a swelling and obliteration of the buccal vestibule.



Figure 2: Panoramic radiograph reveals a transmigrated canine of the right side and the retained deciduous canine.



Figure 3: Intraoral photograph shows the canine that was extracted and the enucleated lesion.

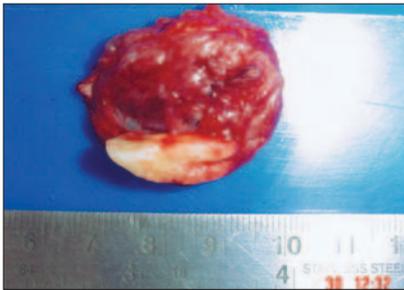


Figure 4: The tooth and the cyst lining after enucleation of the cyst.

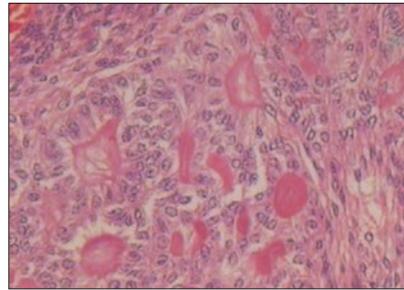


Figure 5: Histopathological slide shows features typical of adenomatoid odontogenic tumour, including sheets of polygonal cells throughout the fibrous connective tissue stroma.

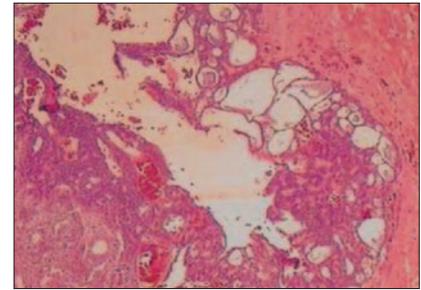


Figure 6: Histopathological slide shows the ductal lumina were surrounded by columnar epithelial cells and filled in some areas with eosinophilic material.

Case Report

A 17-year-old girl presented to the department of dental surgery, All India Institute of Medical Sciences, with swelling and discharge of pus in the lower right canine region. Intraoral examination showed that the labial vestibule was obliterated by expansion of the buccal cortical plate from the lower right canine to the left first premolar region (Fig. 1). The mandibular incisors were displaced labially; the right mandibular deciduous canine was retained but the permanent canine was missing.

A panoramic radiograph revealed a large, well-circumscribed radiolucency extending throughout the anterior mandible from the right canine to the left first premolar region (Fig. 2). The lesion produced an expansion of tissue and extended into the alveolar processes, disrupting the usual orientation of the anterior teeth. The right mandibular canine, which was present within the lesion, had been displaced considerably toward the contralateral side.

On the basis of the clinical and radiographic findings, the differential diagnosis was adenomatoid odontogenic tumour, ameloblastic fibrous odontoma, calcifying odontogenic cyst, calcifying epithelial odontogenic tumour and infected dentigerous cyst.

The patient underwent surgery with local anesthesia. A mucoperiosteal flap from the right to the left premolar region was reflected to expose the labial aspect of the tumour. The labial cortex was very thin and had several areas of complete resorption. The tumour was enucleated along with

the impacted lower permanent canine (Figs. 3 and 4). The areas between the roots of the involved teeth were curetted well. The cavity was packed with Gelfoam (Pharmacia & Upjohn Co, Kalamazoo, Mich.) and the flap was sutured in place. Healing was uneventful, and there was no evidence of recurrence 1 year after the surgery.

Histopathological examination revealed sheets of polygonal cells throughout the fibrous connective tissue stroma (Fig. 5). The ductal lumina were surrounded by columnar epithelial cells and filled in some areas with eosinophilic material (Fig. 6). In other places amorphous calcified material was present. The histopathological report confirmed the diagnosis of adenomatoid odontogenic tumour.

Discussion

Adenomatoid odontogenic tumour is a slowly growing lesion, with a predilection for the anterior maxilla (ratio of cases 2:1 relative to mandible) of young females. Sixty-nine percent of adenomatoid odontogenic tumours are diagnosed in the second decade of life, and more than half occur during the teenage years. The female to male ratio for all age groups and all variants is close to 2:1.¹⁰ Generally the tumours do not exceed 1–3 cm in greatest diameter, but they can be larger, as in the case reported here.⁶ The lesions are typically asymptomatic, but growth of the types with central lesion results in cortical expansion, as in the case reported here. The involved teeth are commonly impacted, and adjacent teeth may be slightly displaced.¹¹ The distribution of unerupted teeth

associated with the follicular type has a typical pattern. The 4 canines combined account for 59% of cases and the maxillary canines alone for 40%.¹² Unerupted first and second molars are rarely involved, nor are deciduous teeth. Root resorption is a less common finding. If the lesion is large, it can cause a painless hard swelling, as in the case reported here.

Adenomatoid odontogenic tumours, accounting for approximately 3% of all odontogenic tumours, are less frequent than odontoma, cementoma, myxoma and ameloblastoma. It has been suggested that this tumour may be a hamartoma rather than a true neoplasm,⁷ but there is currently no evidence to resolve this dispute. A diagnosis of adenomatoid odontogenic tumour should be considered in the differential diagnosis of corticate radiolucencies with small radiopaque foci, especially in teenagers and young adults. For cases in which the lesion appears to surround an unerupted tooth and has no radiopaque component, dentigerous cyst may also be considered in the differential diagnosis. However, an adenomatoid odontogenic tumour often appears to envelop the crown as well as the root, whereas dentigerous cysts do not envelop the roots.¹³⁻¹⁶

The origin of adenomatoid odontogenic tumours is controversial.¹⁷⁻²² Some believe they originate from the odontogenic epithelium of a dentigerous cyst. In addition to the anterior maxilla, the tumour has been reported in other areas of the jaw, such as the angle of the mandible. Therefore, dental lamina remnants likely represent the progenitor cells for this benign odontogenic tumour. According to this hypothesis, the lesion grows (sometimes while forming a cystic space) next to or into a nearby dental follicle, leading to the "envelopmental theory."¹⁷ In the case reported here, the lesion surrounded a fully formed canine tooth, which suggests "envelopmental" pathogenesis. Recent reports indicate that the cells of an adenomatoid odontogenic tumour usually differentiate toward an apparent ameloblastic phenotype but fail to achieve further functional maturation.²²

WHO²³ has described the histologic features of the tumour as follows: "A tumor of odontogenic epithelium with duct like structures and with varying degree of inductive changes 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." The histologic appearance of all variants is identical and exhibits remarkable consistency.^{21,24} At low magnification the most striking pattern is that of various sizes of solid nodules of columnar or cuboidal epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue. Between the epithelial cells of the nodules and in the centre of the rosette-like configuration is found eosinophilic amorphous material, often described as tumour deposits. Conspicuous within the cellular areas are structures of tubular or duct-like appearance. A third characteristic cellular pattern consists of nodules of polyhedral, eosinophilic epithelial cells with squamous appearance and exhibiting well-defined cell boundaries and prominent intracellular bridges. These islands may contain pools of amorphous amyloid-like material and

globular masses of calcified material (thus the suggestion of a combination of calcifying epithelial odontogenic tumour and adenomatoid odontogenic tumour).²⁵ Another epithelial pattern has a trabecular or cribriform configuration. Occasional foci of mitotic activity can be traced. Induction of hyaline, dysplastic dentinoid material or calcified osteodentin has been described. Ultrastructurally, 3 tumour epithelial cell types have been recognized, corresponding to the types that are evident on light microscopy. The connective tissue stroma is very loosely structured and contains thin-walled congested vessels characteristically showing marked degenerative (fibrinoid) changes of the endothelial lining, vessel wall and perivascular connective tissue. It has been suggested recently that the tumour droplets represent some form of enamel matrix.²⁵

Immunohistochemical studies of the lesion suggest expression of keratin and vimentin in the tumour cells at the periphery of the ductal, tubular or whorled structures.²⁷ Amelogenin and enamelin in small mineralized foci are found in the tumour cells and in hyaline droplets.²⁸

Since all variants show identical benign biological behaviour and almost all are encapsulated, conservative surgical enucleation or curettage is the treatment of choice. Recurrence has been reported in very few cases.²⁰ If the follicle is found during surgery to be uninvolved, it can be easily separated from the tumour; it may then be possible to remove the lesion while leaving the teeth in place, as described by Toida and others.²⁹ This would be especially desirable in the maxillary canine region of a young person. However, in the case reported here, the tooth had been pushed to the contralateral side across the midline, and the large size of the lesion justified the decision to remove a tooth along with the lesion. ♦



Dr. Batra is a former senior resident, All India Institute of Medical Sciences, New Delhi. He is currently a clinical research fellow (Commonwealth Scholar), maxillofacial and dental department, Great Ormond Street Hospital for Children, NHS Trust, London, UK.

Dr. Prasad is a former senior resident, All India Institute of Medical Sciences, New Delhi. He is currently a senior house officer (maxillofacial surgery), maxillofacial and dental department, Louisiana State University, Baton Rouge, Louisiana.

Dr. Hari Parkash is professor and head of the department of dental surgery, All India Institute of Medical Sciences, New Delhi.

Correspondence to: Dr. Puneet Batra, Maxillofacial and Dental Department, Great Ormond Street Hospital for Children, NHS Trust, London, UK. E-mail: pbatra87@hotmail.com.

The authors have no declared financial interests in any company manufacturing the types of products mentioned in this article.

References

1. Lucas RB. Pathology of tumors of the oral tissues. 4th ed. Edinburgh, Scotland: Churchill Livingstone; 1984. p. 66.
2. Stafne EC. Epithelial tumors associated with developmental cysts of the maxilla: report of 3 cases. *Oral Surg Oral Med Oral Pathol* 1948; 1:887.

3. Mallon HL, Sabes WR, Monaco F. Ameloblastic adenomatoid tumor. *Oral Surg Oral Med Oral Pathol* 1968; 25:143.
4. Philipsen HP, Birn H. The adenomatoid odontogenic tumor, ameloblastic adenomatoid tumor or adeno-ameloblastoma. *Acta Pathol Microbiol Scand* 1969; 75(3):375-98.
5. Kramer IRH, Pindborg JJ. WHO International histological classification of tumors. No 5. Histological typing of odontogenic tumors, jaw cysts, and allied lesions. Berlin: Springer Verlag; 1971.
6. 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(4):149-58.
7. Regezi JA, Kerr DA, Courtney RM. Odontogenic tumors: analysis of 706 cases. *J Oral Surg* 1978; 36(10):771-8.
8. Courtney RM, Kerr DA. The odontogenic adenomatoid tumor. A comprehensive study of twenty new cases. *Oral Surg Oral Med Oral Pathol* 1975; 39(3):424-35.
9. Dare A, Yamaguchi A, Yoshiki S, Okano T. Limitations of panoramic radiography in diagnosing adenomatoid odontogenic tumor. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1994; 77(6):662-8.
10. 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(2):246-8.
11. Geist SY, Mallon HL. Adenomatoid odontogenic tumor: report of an unusually large lesion in the mandible. *J Oral Maxillofac Surg* 1995; 53(6):714-7.
12. Abrams AM, Melrose RJ, Howell FV. Adenoameloblastoma: a clinical pathological study of ten new cases. *Cancer* 1968; 22(1):175-85.
13. Tsaknis PJ, Carpenter WM, Shade NL. Odontogenic adenomatoid tumor: report of a case and review of the literature. *J Oral Surg* 1977; 35(2):146-9.
14. Bedrick AE, Solomon MP. The adenomatoid odontogenic tumor: an unusual presentation. *Oral Surg Oral Med Oral Pathol* 1979; 48(2):143-5.
15. Curran AE, Miller EJ, Murrah VA. Adenomatoid odontogenic tumor presenting as periapical disease. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997; 84(5):557-60.
16. Lee JK, Lee KB, Hwang BN. Adenomatoid odontogenic tumor: a case report. *J Oral Maxillofac Surg* 2000; 58(10):1161-4.
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(8):348-52.
18. Giansanti JS, Someren A, Waldron CA. Odontogenic adenomatoid tumor (adenoameloblastoma). Survey of 3 cases. *Oral Surg Oral Med Oral Pathol* 1970; 30(1):69.
19. 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(2):190-3.
20. Philipsen HP, Reichart PA. The adenomatoid odontogenic tumor: ultrastructure of tumor cells and non-calcified amorphous masses. *J Oral Pathol Med* 1996; 25(9):491-6.
21. Philipsen HP, Reichart PA, Nikai H. The adenomatoid odontogenic tumor (AOT): an update. *J Oral Pathol Med* 1997; 2:55-60.
22. Philipsen HP, Reichart PA. Adenomatoid odontogenic tumour: facts and figures. *Oral Oncol* 1998; 35(2):125-31.
23. Kramer IRH, Pindborg JJ, Shear M. WHO International histological classification of tumours. Histological typing of odontogenic tumors. 2nd ed. Berlin: Springer Verlag; 1992.
24. Mosqueda-Taylor A, Ledesma-Montes C, Caballero-Sandoval S, Portilla-Robertson J, Ruiz-Godoy Rivera LM, Meneses-Garcia A. Odontogenic tumors in Mexico. A collaborative retrospective study of 349 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997; 84(6):672-5.
25. Takeda Y, Kudo K. Adenomatoid odontogenic tumor associated with calcifying epithelial odontogenic tumor. *Int J Oral Maxillofac Surg* 1986; 15(4):469-73.
26. Ide F, Kusama K. Adenomatoid odontoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002; 94(2):149-50.
27. Tatemoto Y, Tanaka T, Okada Y, Mori M. Adenomatoid odontogenic tumor: co-expression of keratin and vimentin. *Virchows Arch A Pathol Anat Histopathol* 1988; 413(4):341-7.
28. Saku T, Okabe H, Shimokawa H. Immunohistochemical demonstration of enamel proteins in odontogenic tumors. *J Oral Pathol Med* 1992; 21(3):113-9.
29. Toida M, Hyodo I, Okuda T, Tatetsu N. Adenomatoid odontogenic tumor: report of two cases and survey of 126 cases in Japan. *J Oral Maxillofac Surg* 1990; 48(4):404-8.