Dentomaxillofacial Radiology (2012) 41, 342–348

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CASE REPORT

Bilateral adenomatoid odontogenic tumour of the maxilla in a 2-year-old female—the report of a rare case and review of the literature

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Multifocal odontogenic lesions are uncommon and have only been observed in conditions associated with known genetic mutations. To the best of our knowledge, only two cases of multifocal adenomatoid odontogenic tumours (AOT) have previously been reported in the literature. In this study, we report the first case of a bilateral, separate AOT in the maxilla in the midline in a 2-year-old female. The patient presented with bilateral expansile masses in the maxilla on either side of the midline which had been present for 6 months. She was asymptomatic and had occasional difficulty in breathing. The tumour was diagnosed as AOT and was surgically enucleated along with the associated teeth. The patient recovered well and has been on recall for 5 years. The follow-up panoramic radiograph made a fortnight ago revealed evidence of three new radio-opaque lesions with an associated tooth in the region of the anterior mandible, the premolar region of the right maxilla and the molar region of the left maxilla. To acquire additional information about AOT, all reports regarding AOT cited in 'PubMed' from 1995 onward were reviewed and the incidence, clinical features, radiographic features and management of AOT are discussed in this study.
Dentomaxillofacial Radiology (2012) **41,** 342–348. doi: 10.1259/dmfr/63978332

Keywords: adenomatoid odontogenic tumour; odontogenic tumour; maxilla

Introduction

Adenomatoid odontogenic tumour (AOT) is a slow-growing, well-circumscribed tumour representing 3–7% of all odontogenic tumours.[1] Although the tumour is considered rare in the literature, Philipsen et al[2] reported that the tumour ranks fourth among the odontogenic tumours. It was also known as adeno-ameloblastoma, ameloblastic adenomatoid tumour etc, but the World Health Organization (WHO) classification of 1971 adopted the term AOT, which was coined by Philipsen and Brin.[3] In the latest edition of WHO classification of odontogenic tumours in 2005, AOT is classified into the first group of tumours (odontogenic epithelium without ectomesenchyme) instead of the second group (odontogenic epithelium with ectomesenchyme).[4] Because of the absence of ectomesenchyme in immunohistochemical staining and dysplastic dentine, AOT is now considered the result of a metaplastic process rather than epithelial-ectomesenchyme interaction.[4] Evidence suggests that follicular AOTs arise from the reduced enamel epithelium (REE) which lines the follicles of unerupted teeth.[5] Crivelini et al[6] detected the expression of Cytokeratin 14 in AOT, which is also expressed by REE, and concluded that AOT probably originates from REE.[6] The origin of the extrafollicular variant is still not clear;[5] however, Philipsen et al[7] argued that all AOT variants show identical histology and therefore it points towards a common origin from the dental lamina or its remains.

AOT commonly affects the anterior portion of the jaws, especially the maxilla in females.[1] Philipsen and Reichart et al[8] reported that 64.3% of cases occur in the maxilla and commonly involve the canine tooth. In the mandible the involvement of the canine is more common;[8] however, a few cases associated with the embedded third molars have been reported in the literature. Philipsen et al[2] also reported that the involvement of all the four canines is 60.1% and the maxillary canine alone is 41.7%. AOT is commonly seen in the first and second

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Received 29 July 2010; revised 12 October 2010; accepted 4 November 2010

decade of life and is usually asymptomatic.[2] Clinically the patient may complain of an asymptomatic, slow-growing swelling or a missing tooth.[1]

Multifocal AOT is rare and this paper describes the case of a 6-month-old bilateral separate AOT of the anterior maxilla in the midline in a 2-year-old Indian female which was surgically enucleated along with the associated teeth. The patient recovered well and has been on recall for 5 years. The follow-up panoramic radiograph taken a fortnight ago revealed evidence of three new radio-opaque lesions with an associated tooth in the region of the anterior mandible, premolar region of the right maxilla and molar region of the left maxilla, which are yet to be diagnosed.

Case report

A 2-year-old Indian female presented with 6-month-old bilateral expansile masses in the maxilla to our hospital 5 years ago. The patient's mother first noticed a diffuse swelling in the right posterior region of the maxilla followed by involvement of the anterior and the left posterior regions of the maxilla. The swelling had been gradually increasing in size since then and was more pronounced on the right side. The patient had remained asymptomatic with occasional difficulty in breathing. The medical and dental histories were insignificant; the patient's older sister, aged about 9 years, was surgically treated for ameloblastic fibroma of the maxilla at the age of 2 years.

On examination, bilateral expansion of the maxilla was seen and was more pronounced on the right side, extending from the infraorbital rim to the corner of the mouth and laterally 3 cm and 2 cm on the right and the left sides (Figure 1). The skin over the swelling appeared normal with a smooth surface. A narrowing of the airway when breathing through the mouth was evident. On intraoral examination, two diffuse, bony hard swellings were seen in the maxilla on either side of the midpalatine raphe, extending from the incisor teeth to the distal aspect of teeth 55 and 65, with obliteration of the labial and the buccal vestibule bilaterally (Figure 1). The full set of deciduous teeth was seen with spacing and was firm and non-tender. The mucosa over the swelling appeared normal.

The patient did not co-operate for a panoramic radiograph, but an occipitomental radiograph revealed obliteration of the maxillary sinus bilaterally. A CT scan revealed large, well-defined separate expansile mass lesions in both the maxillae, with the lesion on the right side larger than the left and comprising a calcified mass, displaced permanent teeth and fluid. Inferiorly the mass extended to the alveolar ridge; superiorly it extended to the floor of the orbit, resulting in bowing of the orbital floor, while medially the lesions were seen to abut each other inferiorly and the nasal septum superiorly. The right-sided lesion showed posterior cortical destruction with mild extension of the lesion into the soft tissues (Figures 2 and 3). Complete blood count, serum calcium and serum alkaline phosphotase levels were assessed and were well within the normal range. Incisional biopsy from both tumour masses revealed similar findings showing sheets of spindly fibroblastic cells with a few gland-like spaces containing hyaline eosinophilic material. The lining cells were tall and columnar and their nuclei were polarized away from the central lesion. Also seen scattered throughout the lesions were calcified and ossified structures, which were suggestive of AOT (Figure 4).

The diagnosis of AOT bilaterally involving the maxillae was made. The bilateral tumour was surgically enucleated along with the associated deciduous teeth and permanent tooth buds simultaneously under general anaesthesia. The patient recovered well within a week and has been on routine biannual clinical and radiographic follow-up for 5 years (Figure 5). The

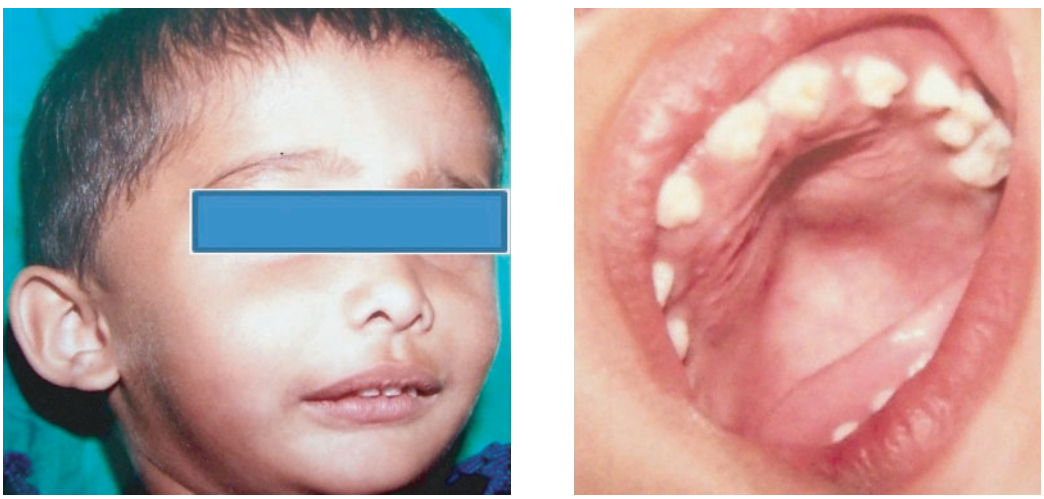

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Figure 1 Bilateral expansile masses in the maxilla, which is more pronounced on the right side

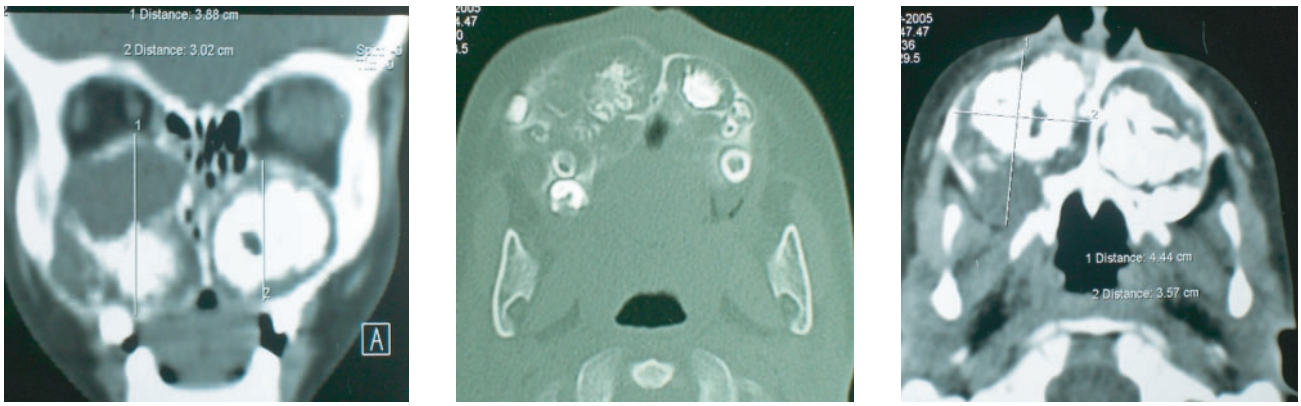

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Figure 2 CT scan revealing the bilateral tumour mass with a calcification and associated tooth in the maxilla. The tumour on the right side measured $4.44 \times 3.57 \times 3.88$ cm and tumour on the left side measured $3.51 \times 2.90 \times 3.02$ cm

follow-up panoramic radiograph taken a fortnight ago revealed evidence of three new radio-opaque lesions associated with a developing tooth bud in the anterior mandible, the premolar region of the right maxilla and the molar region of the left maxilla (Figure 6). The lesions appear radio-opaque with a few radiolucent areas in between surrounded by a radiolucent rim and radio-opaque border, and are associated with developing tooth buds of 31 (20×10 mm), 14 (15 mm diameter) and 27 (15×10 mm). The patient is now on a monthly radiographic follow-up.

Discussion

From 1995 onwards, 76 single cases of AOT (excluding case series of more than 5 cases) have been published in 'PubMed'. The age range was 3–46 years; however, most of the cases reported were in the age group of 10–22 years with a female predominance. Very few cases have been reported in the age range of 1–10 years; the youngest patient was a 3-year-old female[9] and another case was in a 4-year-old female,[10] both of whom had peripheral AOTs. Most of the cases of AOT were predominantly found in the anterior maxilla, followed by the anterior mandible, and a few cases reported AOTs in the posterior mandible in the third molar region.

AOT is commonly seen in the age group 10–19 years.[2] Philipsen and Reichart et al[11] report that two-thirds of the cases are diagnosed in the second decade of life and more than half of the cases occur in the teens (13–19 years). The age range in our literature review was 3–46 years and the case reported here is in a 2-year-old female with central AOT, which to the best of our knowledge is the first case report of AOT in such a young child. There is a predilection of AOT in females (male-to-female ratio = 1:1.9),[8] with reports of higher incidence in females in Asian populations, especially in Sri Lanka

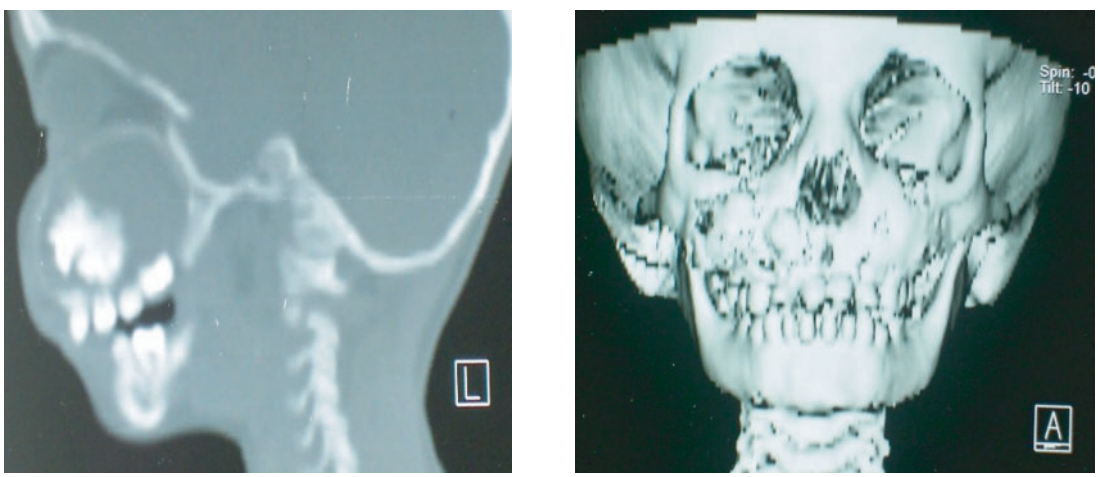

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Figure 3 Sagittal CT scan revealing the large tumour mass with a calcification and a three-dimensional CT showing cortical destruction in the right maxilla

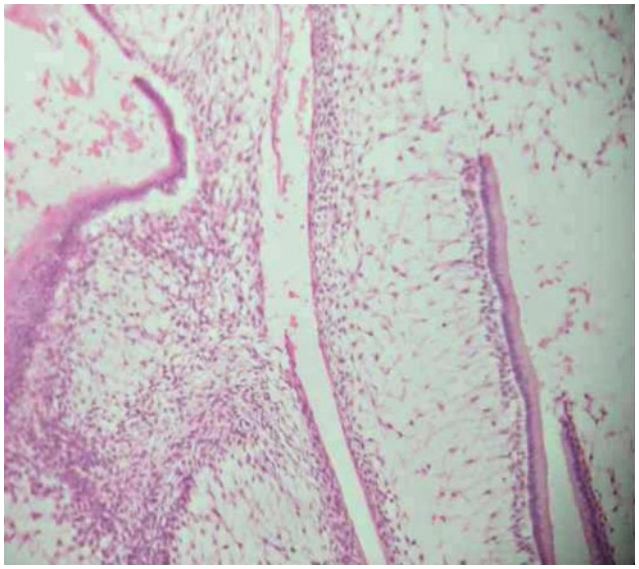

Figure 4 Histopathological picture revealing sheets of spindly fibroblastic cells

(male-to-female ratio = 1:3.2)[12] and Japan (male-to-female ratio = 1:3).[13]

The literature review by the authors revealed cases of AOT arising/associated with dentigerous cyst, calcifying epithelial odontogenic tumour, calcifying odontogenic cyst, odontoma and unicystic ameloblastoma. There are a few reports of cases of adenoameloblastoma with dentinoid which has features of ameloblastoma and AOT (Table 1). Vargas et al[28] reported two cases of a new odontogenic lesion for which they coined the term adenomatoid odontogenic hamartoma, following which a few cases have been reported (Table 1). These tumours are composed of mature hard and soft dental tissue, which resembles a developing tooth, and the interspersed remnants of odontogenic epithelium which form duct-like structures.

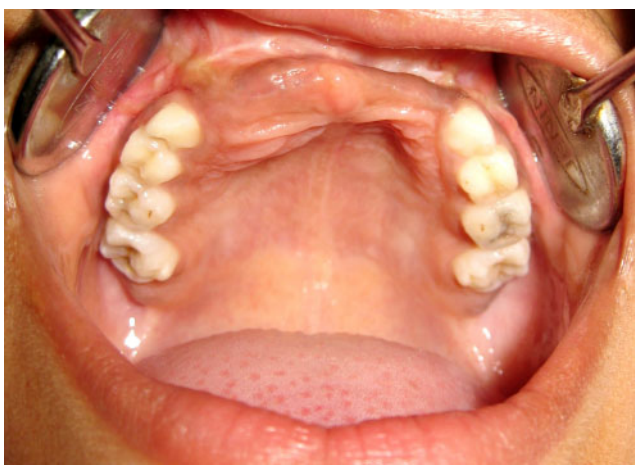

Figure 5 Fifth year follow-up pictures revealing missing anterior teeth from canine to canine in the maxilla and a normal appearing palate

Clinically the patient may present with a missing tooth, especially a canine or an asymptomatic slow growing swelling of the jaw bones.[1] According to Philipsen et al,[2] AOT appears in three clinical subtypes—follicular, extrafollicular and peripheral. The follicular and extrafollicular variants are both intrabony and account for approximately 97% of all AOTs, of which 73% are of a follicular type. The follicular type appears as a unilocular cystic radiolucency associated with an unerupted or impacted tooth and is diagnosed earlier in life (mean age 17 years) than the extrafollicular type (mean age 24 years).[2] It is impossible to distinguish the follicular type of AOT from the dentigerous cyst radiographically.[1] The radiolucency may sometimes extend apically along the root past the cementoenamel junction and this may help in distinguishing AOT from a dentigerous cyst.[1] The extrafollicular type (24%) is a central lesion not associated with a tooth[2] and may present as radiolucent lesions in the periapical region of the tooth mimicking a periapical cyst,[30] or it may present as a periodontal intrabony defect[31] or in the maxillary sinus.[32] The peripheral type is seen as small sessile masses on the facia gingiva of the maxilla, which may mimic gingival epulis or fibroma.[1] Displacement of neighbouring teeth owing to tumour expansion is much more common than root resorption.[33] The peripheral lesion may show some erosion of the adjacent cortical bone.[33] The central tumours produce a corticated radiolucency, sometimes with radio-opaque specks, and in 78% of the cases the radiolucency shows discrete foci and a flocculent pattern of scattered radio-opacities,[34,35] which may mimic the radiographic picture of a calcifying epithelial odontogenic tumour or calcifying odontogenic cyst.

The lesions of AOT are small, seldom exceeding 3 cm in diameter, and are asymptomatic so are discovered during routine radiographic examination, but some larger lesions cause painless expansion of the bone.[1] Tsaknis et al[36] reported a case of massive AOT measuring 12 cm in an 11-year-old female, with seven impacted teeth and a pronounced area of radio-opaqueness. Takahashi et al[37] have reported a gigantic AOT of the maxillary sinus and Geist et al[38] have reported a case of unusually large AOT of the mandible which had caused expansion and resorption of the lower border of the mandible and displacement of the anterior teeth and extended across the midline. In the case reported here the tumour was bilateral and measured about 4 cm on the left side and 4.5 cm on the right side with a calcified mass, displaced permanent teeth and fluid. It had caused expansion of the maxilla, bowing of the orbital floor, posterior cortical destruction on the right side and also mild extension into the soft tissues.

Multifocal odontogenic lesions are uncommon with few case reports in the literature. Sedghizadeh et al[39] have reported a case of a multifocal calcifying epithelial odontogenic tumour, Abrahao et al[40] have reported a case of a recurrent bilateral gingival peripheral calcifying epithelial odontogenic tumour, Kamal et al[41]

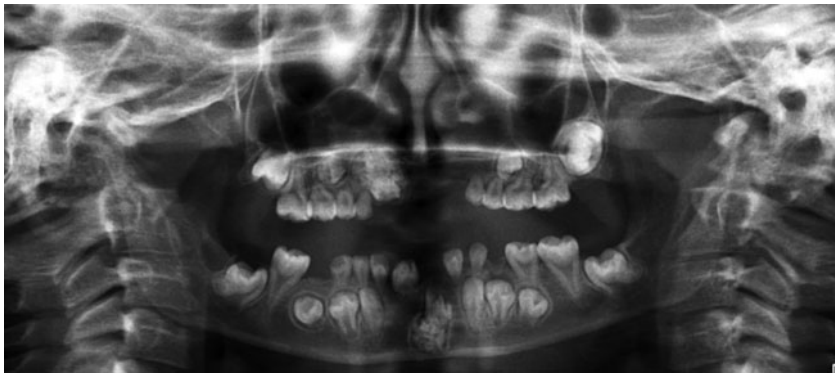

Figure 6 Fifth year panoramic radiograph revealing a radio-opaque area with radiolucency in between, surrounded by a radiolucent rim and radio-opaque border which are associated with the tooth buds of 31 (20 × 10 mm), 14 (15 mm diameter) and 27 (15 × 10 mm)

have reported a case of multifocal peripheral odontogenic fibroma, Straith et al[42] have reported a case of bilateral odontoma, Mills et al[43] have reported a case of a squamous odontogenic tumour and there are a number of reports of multiple keratocystic odontogenic tumours in basal cell bifid rib syndrome in the literature.

Only two cases of multifocal AOTs have been reported in the literature to the best of our knowledge, and this is the first case of a bilateral, separate, multifocal, central AOT in the maxilla in the midline in a 2-year-old female. The follow-up radiographs have also revealed evidence of three new radio-opaque lesions with an associated tooth bud in the region of the anterior mandible, the premolar region of the right maxilla and the molar region of the left maxilla, which may represent new foci of AOT and are yet to be diagnosed. Larsson et al[44] have reported a case of multifocal AOT of the jaw bone in a 12-year-old female who developed a dozen separate radiolucent lesions over a 5-year period. They were removed surgically along with about 20 associated tooth germs and unerupted malformed teeth. Bartake et al[45] have reported a case of two AOTs of the left maxilla in the region of teeth 21–26 in a 14-year-old female which were found to be two separate, well-encapsulated masses separated by thin bony septae during enucleation. Otero et al[29] have reported the first case of a bilateral adenomatoid odontogenic hamartoma in a 12-year-old female, wherein the lesions were well-circumscribed, unilocular radiolucent areas in the mandibular third molar region.

Microscopically an intracystic epithelial proliferation is composed of polyhedral to spindle cells.[1] The pattern is typically lobular, although some areas may show a syncytial arrangement of cells.[1] Rosettes and duct-like structures of columnar epithelial cells containing small amounts of eosinophilic material give the lesion its characteristic microscopic features, but may be scanty or even absent in a given lesion.[1] Small deposits of calcified material are scattered over a background of odontogenic cells.[1]

AOT is a well-encapsulated tumour and is benign so it can be surgically enucleated easily from the bone along with the capsule.[1] Recurrence of the tumour or aggressive behaviour are generally not the features of AOT, although Chuan-Xiang et al[46] reported a case where recurrence appeared twice in a 36-year-old male over a 20-year period. Garg et al[47] also reported the case of a 20-year-old female with large, aggressive AOT which caused resorption of roots and Takigami et al[48] reported a similarly aggressive AOT which recurred four times and extended to the base of the skull. The above case reports describe recurrence of AOT with features of aggressiveness, root resorption and rapid growth, which is generally not the behaviour of AOT and the diagnoses in these cases can be questioned.

In the case reported here the large, bilateral, simultaneous occurrence of AOT in the midline of the maxilla in a 2-year-old child makes it a very rare presentation of this tumour. The patient recovered well after the enucleation and there has been no recurrence of the tumour until now. The latest follow-up radiographs have revealed evidence of three new radio-opaque lesions with an associated tooth bud in the region of the anterior mandible, the premolar region of the right maxilla and the molar region of the left maxilla which may represent new foci of AOT and are yet to be diagnosed.

Table 1 Adenomatoid odontogenic tumours are thought to arise from/associated with as reported in the literature

Tumours	*Cases*	*Authors*
Dentigerous cyst	Two	Nonaka et al,[14] Bravo et al[15]
Dental cyst	One	Gracia-Pola Vallejo et al[16]
Odontoma	Two	Cudney et al,[17] Martinez et al[18]
Adenoameloblastoma with dentinoid	Six	Ghasemi-Moridani and Yazdi,[19] Evans et al,[20] Allen et al[21]
Calcifying epithelial odontogenic tumour	Two	Mosqueda-Taylor et al,[22] Miyake et al[23]
Calcifying odontogenic cyst	Three	Phillips et al,[24] Buch et al,[25] Zeitoun et al[26]
Unicystic ameloblastoma	Three	Jivan et al,[5] Raubenheimer et al[27]
Adenomatoid odontogenic hamartoma	Three	Vargas et al,[28] Otero et al[29]

References

1. Neville BW, Damm DD, Allen CM, Bouquet JE. *Oral and maxillofacial pathology*, 2nd edn. WB Saunders: Philadelphia, PA; 2002. pp. 621–623.
2. Philipsen HP, Reichart PA, Zhang KH, Nikai H, Yu QX. Adenomatoid odontogenic tumour: biologic profile based on 499 cases. *J Oral Pathol Med* 1991; **20**: 149–158.
3. Philipsen HP, Brin H. The adenomatoid odontogenic tumour, ameloblastic adenomatoid tumour or adeno-ameloblastoma. *Acta Pathol Microbiol Scand* 1969; **75**: 375–398.
4. Jing W, Xuan M, Lin Y, Wu L, Liu L, Zheng X, et al. Odontogenic tumours: a retrospective study of 1642 cases in a Chinese population. *Int J Oral Maxillofac Surg* 2007; **36**: 20–25.
5. Jivan V, Altini M, Meer S, Mahomed F. Adenomatoid odontogenic tumour (AOT) originating in a unicystic ameloblastoma: a case report. *Head Neck Pathol* 2007; **1**: 146–149.
6. Crivelini MM, de Araujo VC, de Sousa SO, de Araujo NS. Cytokeratins in epithelia of odontogenic neoplasms. *Oral Dis* 2003; **9**: 1–6.
7. Philipsen HP, Samman N, Ormiston IW, Wu PC, Reichart PA. Variants of the adenomatoid odontogenic tumour with a note on tumour origin. *J Oral Pathol Med* 1992; **21**: 348–352.
8. Philipsen HP, Reichart PA, Siar CH, Ng KH, Lau SH, Zhang X, et al. An updated clinical and epidemiological profile of the adenomatoid odontogenic tumour: a collaborative retrospective study. *J Oral Pathol Med* 2007; **36**: 383–393.
9. Kearns GJ, Smith R. Adenomatoid odontogenic tumour: an unusual cause of gingival swelling in a three year old patient. *Br Dent J* 1996; **181**: 380–382.
10. Unal T, Cetingul E, Gunbay T. Peripheral adenomatoid odontogenic tumour: birth of a term. *J Clin Pediatr Dent* 1995; **19**: 139–142.
11. Philipsen HP, Reichart PA. Adenomatoid odontogenic tumour: facts and figures. *Oral Oncol* 1999; **35**: 125–131.
12. Nigam S, Gupta SK, Chaturvedi KU. Adenomatoid odontogenic tumour – a rare cause of jaw swelling. *Braz Dent J* 2005; **16**: 251–253.
13. Sato D, Matsuzaka K, Yama M, Kakizawa T, Inoue T. Adenomatoid odontogenic tumour arising from the mandibular molar region: a case report and review of literature. *Bull Tokyo Dent Coll* 2004; **45**: 223–227.
14. Nonaka CFW, de Souza LB, Quindere LB. Adenomatoid odontogenic tumor associated with dentigerous cyst—unusual case report. *Braz J Otorhinolaryngol* 2007; **73**: 129–131.
15. Bravo M, White D, Miles L, Cotton R. Adenomatoid odontogenic tumor mimicking a dentigerous cyst. *Int J Pediatr Otorhinolaryngol* 2005; **69**: 1685–1688.
16. Gracia-Pola Vallejo M, Gonzalez Gracia 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**: 55–58.
17. Cudney N, Persico J, Cordell KG, D'Silva NJ. Adenomatoid odontogenic tumor developing in association with odontoma: report of a case. *Quintessence Int* 2008; **39**: 693–697.
18. Martinez A, Taylor AM, Marchesani FL, Brethauer U, Spencer ML. Adenomatoid odontogenic tumor concomitant with cystic complex odontoma: case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009; **108**: e25–e29.
19. Ghasemi-Mordani S, Yazdi I. Adenoid ameloblastoma with dentinoid: a case report. *Arch Iranian Med* 2008; **11**: 110–112.
20. Evans BL, Carr RF, Phillipe LJ. Adenoid ameloblastoma with dentinoid: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2004; **98**: 583–587.
21. Allen CM, Neville BW, Hammond HL. Adenomatoid dentinoma: report of four cases of unusual odontogenic lesion. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1998; **86**: 313–317.
22. Mosqueda-Taylor A, Carlos-Bregni R, Ledesma-MontesC, Fillipi RZ, de Almeida OP, Vargas PA. Calcifying epithelial odontogenic tumor like areas are common findings in adenomatoid odontogenic tumors and not a specific entity. *Oral Oncol* 2005; **41**: 214–215.
23. Miyake M, Nagahata S, Nishihara J, Ohbayashi Y. Combined adenomatoid odontogenic tumor and calcifying epithelial odontogenic tumor: report of case and ultrastructural study. *J Oral Maxillofac Surg* 1996; **54**: 788–793.
24. Philips MD, Closmann JJ, Baus MK, Torske KR, Williams SB. Hybrid odontogenic tumor with features of ameloblastic fibro-odontoma, calcifying odontogenic cyst, and adenomatoid odontogenic tumor: a case report and review of literature. *J Oral Maxillofac Surg* 2010; **68**: 470–474.
25. Buch RS, Coerdt W, Wahlmann U. Adenomatoid odontogenic tumor in calcifying odontogenic cyst. [In German.] *Mund Kiefer Gesichtschir* 2003; **7**: 301–305.
26. Zeitoun IM, Dhanrajani PJ, Adeyemi Mosandomi H. Adenomatoid odontogenic tumor arising in calcifying odontogenic cyst. *J Oral Maxillofac Surg* 1996; **54**: 634–637.
27. Raubenheimer EJ, Seeliger JE, Van HeerdenWF, Dreyer AF. Adenomatoid odontogenic tumor: a report of two large lesions. *Dentomaxillofac Radiol* 1991; **20**: 43–45.
28. Vargas PA, Carlos-Bregni R, Mosqueda-Taylor A, Cuairan-Ruidiaz V, Lopes MA, de Almeida OP. Adenomatoid dentinoma or adenomatoid odontogenic hamartoma: what is the better term to denominate this uncommon odontogenic lesion. *Oral Dis* 2006; **12**: 200–203.
29. Otero D, Simões M, Antero S, Lourenço S, Niteroi RJ. Bilateral adenomatoid odontogenic hamartoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009; **107**: e24–e26.
30. Philipsen HP, Srisuwan T, Reichart PA, Alcantara SP. Adenomatoid odontogenic tumour mimicking a periapical cyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002; **94**: 246–248.
31. Blumenthal NM, Mostofi R. Repair of an intrabony decfect from an adenomatoid odontogenic tumour. *J Periodontal* 2000; **71**: 1637–1640.
32. Friedrich RE, Scheuer HA, Zustin J. Adenomatoid odontogenic tumour (AOT) of maxillary sinus: case report with respect to immunohistochemical findings. *In Vivo* 2009; **23**: 111–116.
33. Handschel JGK, Depprich RA, Zimmermann AC, Braunstein S, Kübler NR. Adenomatoid odontogenic tumour of the mandible: review of the literature and report of a rare case. *Head Face Med* 2005; **1**: 3.
34. Dare A, Yamaguchi A, Yoshiki S, Okano T. Limitation of panoramic radiography in diagnosing adenomatoid odontogenic tumours. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1994; **77**: 662–668.
35. Toida M, Hyodo I, Okuda T, Tatematsu N. Adenomatoid odontogenic tumour: report of two cases and survey of 126 cases in Japan. *J Oral Maxillofac Surg* 1990; **48**: 404–408.
36. Tsaknis PJ, Carpenter WM, Shade NL. Odontogenic adenomatoid tumour: report of case and review of the literature. *J Oral Surg* 1977; **35**: 146.
37. Takahashi H, Fujita S, Shibata Y, Yamaguchi A. Adenomatoid odontogenic tumour: immunohistochemical demonstration of transferring, ferritin and alpha-one-antitrypsin. *J Oral Pathol Med* 2001; **30**: 237–244.
38. Geist SM, Mallon HL. Adenomatoid odontogenic tumour: report of an unusually large lesion in the mandible. *J Oral Maxillofac Surg* 1995; **53**: 714–717.
39. Sedghizadeh PP, Wong D, Shuler CF, Linz V, Kalmar JR, Allen CM. Multifocal calcifying epithelial odontogenic tumour. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007; **104**: e30–34.
40. Abrahão AC, Camisasca DR, Bonelli BR, Cabral MG, Lourenço SQ, Torres SR, et al. Recurrent bilateral gingival peripheral calcifying epithelial odontogenic tumour (Pindborg tumour): a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009; **108**: e66–71.
41. Kamal R, Palaskar S, Shetty VP, Bhushan A. Multifocal peripheral odontogenic fibroma. *J Oral Maxillofac Pathol* 2008; **12**: 72–74.
42. Straith FE. Bilateral odontoma: report of a case. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* l950; **3**: 1036–1040.

43. Mills WP, Davila MA, Beuttnmuller EA, Koudelka BM. Squamous odontogenic tumour: report of a case with lesions in three quadrants. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1986; **61**: 557–563.
44. 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.
45. Bartake AR, Punnya VA, Sudeendra P, Rekha K. Two adenomatoid odontogenic tumours of the maxilla: a case report. *Br J Oral Maxfac Surg* 2009; **47**: 638–640.
46. Chuan-Xiang Z, Yang G. Adenomatoid odontogenic tumour: a report of a case with recurrence. *J Oral Pathol Med* 2007; **36**: 440–443.
47. Garg D, Palaskar S, Shetty VP, Bhushan A. Adenomatoid odontogenic tumour – hamartoma or true neoplasm: a case report. *J Oral Sci* 2009; **51**: 155–159.
48. Takigami M, Uede T, Imaizumi T, Ohtaki M, Tanabe S, Hashi K, et al. A case of adenomatoid odontogenic tumour with intra-cranial extension. *No Shinkei Geka* 1988; **16**: 775–779.