Case Report

A case report and short review on changing trends in the site of occurrence of adenomatoid odontogenic tumor: Unravelling the past 15 years

Sneha Sethi1, Manish Kumar2, Pratul Aggarwal3, Indra Kumar H.S.4, Chetan D. Sugandhi5, Silvie Singh6

1Department of Oral and Maxillofacial Pathology, Sudha Rustagi College of Dental Sciences and Research, Faridabad, Haryana, 2Departments of Oral and Maxillofacial Pathology, 3Prosthodontics, 4Periodontics and 5Oral Pathology, Tatyasaheb Kore Dental College and Research Centre, Kohlapur, Maharashtra, 6Department of Conservative Dentistry and Endodontics, DJ Dental College, Modinagar, Uttar Pradesh, India

ABSTRACT

Adenomatoid odontogenic tumor (AOT) is an uncommon benign odontogenic lesion, with debatable histogenesis and variable histopathology. A systematic and diverse insight into the evolution, clinical presentation, histology, and immunohistochemical findings of this lesion is reviewed and presented. We reviewed the data published from 2000 to 2014 of approximately 255 cases that revealed a significant change in the incidence of predominant site involved, in contrast to the findings published by Reichart. We have also included the chronological order of events leading to the coining of the term AOT, which shows the curiosity that has been dedicated to understanding the lesion. Immunohistochemistry is considered to be a hallmark in pathology for learning the molecular pathogenesis and giving a correct final diagnosis. Several markers have been used to investigate and understand this lesion, and a compilation of the findings has been tabulated.

Key Words: Ameloblastoma, immunohistochemistry, incidence, odontogenesis

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) was first elucidated by Driebaldt in 1907 as “pseudo-adenoameloblastoma,” and later as “adenomatoid odontogenic tumor.”[1] The World Health Organization (WHO) in 2005 defined AOT as a tumor composed of odontogenic epithelium, presenting a variety of histo-architectural patterns, embedded in mature connective tissue stroma, and characterized by slow and progressive growth.[2]

AOT is a benign, hamartomatous, noninvasive, uncommon, epithelial lesion of the odontogenic origin. It has tendency to affect the younger age group usually during the second decade, also an apparent inclination toward female presentation, as the established male to female ratio of occurrence is 1:2. This lesion is known to be allied with unerupted canines and lateral incisors. The clinical course of the lesion is slow and remains clinically unnoticeable for a long time. The deformity produced by this lesion manifests as displacement of adjoining teeth and an obvious expansion of the surrounding bone.[1,2] Sometimes, it may be also as “two-thirds tumor” because:

- Two-third occurrence in maxilla
- Two-third female preponderance
- Two-third association with unerupted tooth
- Two-third affected teeth are canines.

The lesion when associated with an impacted (maxillary permanent canines account
for 41.7% and all four canines for 60.1% of AOT-associated embedded teeth) and a displaced tooth is referred to as a follicular variant; the origins of this variant are considered to be the reduced enamel epithelium of the dental follicle. It contributes for 73.0–97.2% of all reported cases and is diagnosed earlier in life usually in the second decade.[3,4] When AOT mimics a radicular cyst with manifestation around the apex of a tooth, it is categorized as an extrafollicular variant; the origins of which remain unclear, but it has been suggested that odontogenic cysts or cystic tumors may undergo secondary changes to result in the formation of this benign lesion. Only 12 cases of a rare subvariant have been reported where the lesion impersonates a periapical abscess on radiographic investigation.[3-4] Sometimes, we see a peripheral manifestation which originates at a distance from the tooth germs and is rarely encountered (14 cases). They show a characteristic bone defect or ectopic growth and a significant predominance in females, the maxillary region, especially the anterior maxilla, with primary involvement seen of incisors and sometimes into the maxillary antrum. The histological features of variants of AOT are characteristic and perpetually distinguishable.[3-5]

A decade-long controversial debate on the true classification this tumor has prevailed, hamartoma or neoplasm? The followers of the hamartoma category justify their thinking by pointing to the restricted growth potential and limited sizes along with an absent inherent capacity to reoccur.[10] On the contrary, the supporters of the neoplastic theory argue to by suggesting the lesion is slow growing and early removal prevents its growth to clinically noticeable sizes and also state that many cases which have been left untreated have grown to considerable sizes causing facial asymmetry and distortion.[6] Furthermore, they point out that the spectrum of histologic patterns which are observed in AOT is inconsistent with the variation seen in a developmental anomaly.[8-10] The idea of origin of this lesion from reduced enamel epithelium is enforced by many ultrastructural and immunohistochemical studies along with the resemblance of cytological features seen in this lesion to components of derived from enamel organ; the occurrence in tooth-bearing region of jaws and its unavoidable alliance with impacted teeth has further strengthened this notion. The 1971 WHO classification stated: “It is generally believed that the lesion is not a neoplasm.” However, Handschel et al. concluded that “such a controversy is unresolvable because sound arguments can be advanced in favor of and against both hypotheses. The arguments are based on personal bias rather than on scientific evidence.”[11]

The expansion of specific antibodies for immunohistochemistry has produced substantial growth during the past few years helped us understand the histogenesis of this tumor. A detailed discussion on the immunohistochemical features has been included in the later part of the discussion of this article.

Here, we present an unusual presentation of this lesion in the mandible causing extensive jaw swelling.

**CASE REPORT**

A 15-year-old female patient, with an asymmetrical anterior mandibular swelling, reported to clinics. On examination, the face appeared asymmetrical with a swelling seen in the front region of the lower jaw, approximately 2.5 cm × 3.5 cm in size, extending from the lower lip to 1 cm below the lower border of the mandible. The overlying skin was tense, normal in color with no draining sinuses. The swelling was nontender, noncompressible, nonfluctuant, firm to hard in consistency with diffuse margins. There was no palpable lymphadenopathy, and there was apparent deviation of the jaw to the left side on opening of mouth [Figure 1a].

Intraoral examination revealed the presence of a solitary unilateral swelling in the lower jaw, extending from the distal aspect of central right mandibular incisor; crossing the midline up to left mandibular second premolar region with missing or impacted permanent canine. Superoinferiorly, it extended from the gingival margin obliterating the lower left facial vestibule. The left mandibular canine and premolars were lingually inclined [Figure 1b].

**Radiographic features**

Orthopantomogram showed [Figure 2a] a well-defined unilocular radiolucency extending anteroposteriorly from 31 to mesial aspect of 36 and superoinferiorly from gingival margin/inferior to roots of the inclined canine and from premolars to inferior border of the mandible. Radiopacities in the form of flecks suggestive of calcifications and a single large radiopaque mass suggestive of tooth are appreciable.

Computed tomography revealed an irregular thick cystic lesion with areas of calcification in the left parasymphyseal region [Figure 2b].
Microscopy
Fine needle aspiration cytology from the lesion showed proteinaceous fluid with few red blood cells, polymorphonuclear lymphocytes, and macrophages. No definitive diagnosis could be made.

The microscopic picture of the lesion revealed the presence of a single large cystic space and odontogenic epithelium in scanty connective tissue stroma surrounded by thick fibrous capsule. The odontogenic epithelium is arranged in sheets, duct-like and convoluted/whorled patterns. The ductal patterns are peripherally lined by single layer of ameloblast-like cells with nuclei away from the central space and clear cystic spaces. The convoluted patterns show spindle-shaped cells surrounded by amorphous eosinophilic material [Figure 3].

After carefully analyzing the clinical, radiographic, and histopathological findings, we reached to a final diagnosis of AOT. The patient was referred to the department of oral surgery for excision of lesion. Excision of the lesion from the mandible caused no problems.

A follow-up of 3 and 6 months was recorded; there were no signs of recurrence of the lesion till date. The wound healed uneventfully, and radiographically, no suspicious activity was observed.

DISCUSSION
A review of the literature on AOT was performed, the database used was PubMed interface of MEDLINE, only the case reports with confirmed histopathological diagnosis were included and collision tumors were excluded, the case series reported by authors were also included. The #MeSH words used were adenomatoid odontogenic tumor, odontogenic, and case report. We reviewed the data associated with AOT from 2000 to 2014,[3-8,13-93] and it was found that in the last 14 years, there was a record of 255 reported cases.

Out of 255 cases reviewed, 108 cases belonged to the mandibular anterior quadrant, 52 cases belonged to the mandibular posterior quadrant, 45 cases belonged to the maxillary anterior quadrant, and only 4 cases belonged to the maxillary posterior quadrant [Graph 1]. The age of occurrence of this lesion ranged from 2 to 44 years. An analysis of the mean age was performed separately in each quadrant and found it to be 19.5 years in the maxillary anterior quadrant, 19 years in the maxillary posterior quadrant.
Sethi, et al.: Changing trends in adenomatoid odontogenic tumor

quadrant, 20.5 years in the mandibular anterior quadrant, and 17.8 years in the mandibular posterior quadrant [Graph 2]. Another comparison of site predilection was evaluated in males and females in each quadrant, which showed only 32% of maxillary anterior quadrants were associated with males and 68% were females, whereas in the maxillary posterior quadrant, 44% were males and 56% were females; in the mandibular anterior quadrant, 40.5% were males and 59.5% were females; in the mandibular anterior quadrant, 35% were males and 65% were females [Graph 3]. An overall female predominance was observed (62.12%) in the present study, with a female to male ratio of 1.45:1.

Our data were compared with the comprehensive analysis performed by Reichart and Philipsen[5] and a few striking points were noted [Graph 4]. Out of the 532 cases, 341 were associated with permanent unerupted teeth, in which 209 cases were recorded in the maxillary anterior quadrant (61.2%), 112 cases in the mandibular anterior quadrant (32.3%), 12 cases in the maxillary posterior quadrant (3.51%), and 8 cases in the mandibular posterior quadrant (2.34%). The mean age range recorded by them was 3–82 years; in contrast to our findings, the range of age of occurrence of the lesion to be 2–44 years. The mean ages recorded by both studies were apparently similar. The gender incidence of each decade was calculated and compared by the similar analysis performed by Reichart and Philipsen[5] [Graph 5]. The females and males in the 0–9 years age decade were 3.3% and 1.5% according to Reichart and 4.2% and 2.8% in our study; the female and male incidence in 10–19 year age decade was 43.7% and 24.8% according to Reichart and 47% and 20.2% in the present study; the female and male incidence in 20–29 year age decade was 15.2% and 4.1% according to Reichart and 14.5% and 3.6% in our study; in all cases above 30 years of age, female and male incidence was 2.8% and 4.3% according to Reichart and 3.5% and 4% in the present study.

Various terminologies have been used to describe this lesion and many have been discarded in the process; Table 1 shows the evolution of the term “AOT” in a chronological manner; Table 2 depicts the various terminologies which have been used to describe this lesion.

Microscopically, AOT presents an array of unique and distinctive features. This tumor is almost always delimited by a fibrous capsule which is usually well developed. The primary tumor cells are cuboidal or polygonal epithelial cells, sometimes spindle-shaped cells which are arranged in a characteristic variety of histomorphologic patterns.[7] They can form duct-like spaces with fluctuating diameter, but this pattern is not
predominantly seen. The ducts are lined by a single layer of cuboidal to columnar epithelial cells that have nuclei that frequently are polarized away from the lumen.\[12\] These duct-like structures are frequently lined by an eosinophilic rim of varying thickness called as the hyaline ring. Anastomosing strands of basaloid epithelial cells which resemble cell rests of the dental lamina, arranged in a plexiform, cribriform, trabecular, or lattice-like configuration.\[112\]

Amorphous homogenous material which is eosinophilic (tumor-droplets) is usually seen in the core of these rosettes.\[12\] Furthermore, darkly staining dystrophic calcifications in inconsistent amounts is also a feature observed in the histological examination of these lesions. Other materials which are associated with degraded enamel, mostly, due to a metaplastic process and not an induction incident, such as hyaline, dysplastic, or calcified osteodentin, are other uncommon findings in AOTs.\[2\] The presence of cystic areas in AOTs mimics odontogenic cysts, such as dentigerous cyst, has been reported in the literature (Leon et al., 56.4%).\[45,113\]

These tumors present a minimal mature connective tissue stroma, which is generally loosely structured and contain thin-walled congested vessels with peripheral hyalinization rather apparent. According to el-Labban and Lee,\[114\] an estimate of 70–90% of the blood vessels found in the stroma shows degenerative changes affecting both the endothelial lining and the perivascular connective tissue. These authors attribute these vascular and perivascular changes to the multiplying
basal lamina which is associated with the collagen surrounding the blood vessels undergoing degenerative changes.\cite{114} Philipsen and Reichart emphasize the easy detection of these changes at light microscopic level as they the degeneration is significantly evident.\cite{7} A few cases of AOT have also exhibited melanocytes which can be attributed to the interaction of neural crest cells with developing odontogenic epithelium.\cite{115,116}

Immunohistochemical studies have provided us with confirmatory evidence supporting the odontogenic origin of this lesion [Table 3].\cite{117-126}

- Two distinct varieties of cells (duct and nonduct) have been identified, where none of the enamel matrix proteins such as enamelin, amelogenin, and sheathelin showed positivity by duct forming cells although the nonductal cells were positive for amelogenin\cite{127-130}
- The periluminal and intraluminal material were found to be positive with laminin, type IV collagen, heparan sulfate, proteoglycans, fibronectin, amelogenin, and enamelin\cite{116,127,129,131}
- A cytoplasmic expression of sheathelin has been observed by the cells in the vicinity of the hyaline droplets\cite{129}
- Calcifications were positive for amelogenin, enamelin, and namelysin and negative for sheathelin\cite{128-130,132,133}
- A variety of spindle cells is observed in the intranodular and intermodal spaces and the juxta-tumor spindle cells showed no expression with the enamel matrix proteins similar to ductal cells, suggesting it to be a predecessor of this variety of cells.\cite{134}

**CONCLUSION**

To summarize, we reviewed 255 reported cases of AOT from 2000 to 2014 and observed a striking paradigm shift with respect to prevalence of location.

**Table 3: Immunohistochemical findings by various authors**

| Marker      | Cells (positivity)                                                                 | Author                   |
|-------------|-----------------------------------------------------------------------------------|--------------------------|
| Collagen IV | Basement membrane of cribriform, areas and hyaline materials (+++), epithelial     | Nagatsuka et al.\cite{117}|
|             | whorls, mineralized foci (+)                                                     |                          |
| Versican    | Connective tissue stroma (++), epithelial cells (+)                               | Ito et al.\cite{118}     |
| CK8         | Intense expression                                                               | Larsson et al.\cite{51}  |
| CK5         | Peripheral cells (+)                                                             |                          |
| CK17        | Peripheral cells (+)                                                             |                          |
| CK19        | Peripheral cells (+)                                                             |                          |
| OPG         | (+++), stromal cells                                                              |                          |
| RANKL       | (+++), stromal cells                                                              |                          |
| Integrin α2β1, α3β1, α5β1 | (+) tumor cells                           | de Souza Andrade et al.\cite{120} |
| MMP1        | Stroma and parenchyma (+)                                                         | de Souza Andrade et al.\cite{120} |
| MMP2        | 60% tumor cells (+++), 80% stromal cells (++)                                      | Ribeiro et al.\cite{121} |
| MMP9        | Parenchymal and stromal cells                                                     |                          |
| AE1/AE2     | Superficial (+), ductal (+++), basaloled (+++), fusiform (+++), cyst basal (+++), syst superficial (+++) | Friedrich et al.\cite{19} |
| CK18        | Cyst basal (+), cyst superficial (+)                                             |                          |
| CK14        | Duct like (+++), basaloled (+++), fusiform (+++), cyst basal (+++), cyst superficial (+++) |                          |
| CK5/6       | Duct like (+++), basaloled (+++), fusiform (+++), cyst basal (+++), cyst superficial (+++) |                          |
| CK19        | Superficial (+++), ductal (+++), basaloled (+), cyst basal (+), syst superficial (+++) |                          |
| P63         | Duct like (+++), basaloled (+++), fusiform (+++), cyst basal (+++)                |                          |
| VIMENTIN    | Basaloled (+), fusiform (+)                                                       |                          |
| SMA         | Duct-like (+++)                                                                  |                          |
| EMA         | Superficial (+++), duct like (+), cyst superficial (+++)                          |                          |
| Osteonectin | Epithelial cells                                                                 | Modolo et al.\cite{172} |
| Osteopontin | Calcification foci                                                               | Kumar et al. (2011)\cite{39} |
| Cyclin D1   | Whorls, nuclear stain                                                             | Salehinejad et al.\cite{172} |
| PCNA        | Mild staining all tumor cells                                                    |                          |
| P53         | Mild staining all tumor cells                                                    |                          |
| Podoplanin  | Spindle cells positive                                                           | Tsuneki et al.\cite{124} |
| c-met       | Cytoplasm of epithelial tumor cells                                               | Crivelini et al.\cite{129} |
| HGF         | Cytoplasm of epithelial tumor cells                                               |                          |
| c-myc       | Tumor cells (80%)                                                                | Moosvi et al. (2013)\cite{126} |
| B-catenin   | Tumor cells (cytoplasmic expression – [++] )                                      | Harnet et al.\cite{74}  |
Reichert and Philipsen\textsuperscript{5}) concluded that maxillary anterior area is the most common site for AOT occurrence, whereas our data revealed that in the last 14 years, there is more incidence of AOT in the mandibular anterior quadrant. The origin of AOT is still debatable as to whether AOT is a hamartoma or neoplasm has not been clarified. The lesion has struggled throughout for its name and origin, and an evaluation of the immunohistochemical data also shows us no specific diagnostic marker for this particular odontogenic lesion. Therefore, further detailed studies are required to unveil the secrets of AOT.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or non-financial in this article.

**REFERENCES**

1. Prasad G, Nair P, Thomas S, Gharote H, Singh N, Bhambhal A. Extral follicular adenomatoid odontogenic tumour. BMJ Case Rep 2011;2011. pii: Bcr0320113963.
2. Philipsen HP, Nikal H. Adenomatoid odontogenic tumor. In: Barnes L, Eveson JW, Reichart P, Sidransky D, editors. Pathology and Genetics of Head and Neck Tumors. Lyon: IARC Press; 2005. p. 304-5.
3. Prakasam M, Tiwari S, Satpathy M, Banda VR. Adenomatoid odontogenic tumour. BMJ Case Rep 2013;2013. pii: Bcr2013010212.
4. 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.
5. Reichart PA, Philipsen HP. Odontogenic Tumors and Allied Lesions. United Kingdom: Quintessence Publishing Co. Ltd.; 2004. p. 105-15.
6. Marx RE, Stern D. Oral and Maxillofacial Pathology: A Rationale for Diagnosis and Treatment. Hanover Park: Quintessence Publishing; 2003. p. 609-12.
7. Philipsen HP, Reichart PA. Adenomatoid odontogenic tumour: Facts and figures. Oral Oncol 1999;35:125-31.
8. 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.
9. de Matos FR, Nonaka CF, Pinto LP, de Souza LB, de Almeida Freitas R. Adenomatoid odontogenic tumor: Retrospective study of 15 cases with emphasis on histopathologic features. Head Neck Pathol 2012;6:430-7.
10. Rick GM. Adenomatoid odontogenic tumor. Oral Maxillofac Surg Clin North Am 2004;16:333-54.
11. 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;1:3.
12. 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.
13. Krishnamurthy K, Balaji RS, Devadiga S, Prasad RG. Adenomatoid odontogenic tumor in the maxillary antrum: A rare case entity. J Pharm Bioallied Sci 2014;6 Suppl 1:S196-9.
14. Acharya S, Goyal A, Rattan V, Vaiphei K, Kaur Bhatia S. Dentigerous cyst or adenomatoid odontogenic tumor: Clinical radiological and histopathological dilemma. Case Rep Med 2014;2014:514720.
15. Baskaran P, Misra S, Kumar MS, Mithra R. Adenomatoid odontogenic tumor – A report of two cases with histopathology correlation. J Clin Imaging Sci 2011;1:64.
16. Saluja R, Kaur G, Singh P. Aggressive adenomatoid odontogenic tumor of mandible showing root resorption: A histological case report. Dent Res J (Isfahan) 2013;10:279-82.
17. More CB, Das S, Gupta S, Bhavsar K. Mandibular adenomatoid odontogenic tumor: Radiographic and pathologic correlation. J Nat Sci Biol Med 2013;4:457-62.
18. Shreedhar B, Ali I, Agarwal A, Alam S. A huge adenomatoid odontogenic tumor of maxilla. Case Rep Med 2012;2012:317341.
19. 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.
20. 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.
21. Friedrich RE, Zustin J, Scheuer HA. Adenomatoid odontogenic tumour of the mandible. Anticancer Res 2010;30:1787-92.
22. Ide F. Inter-radicular adenomatoid odontogenic tumor of the anterior mandible. J Oral Maxillofac Surg 2010;68:490-1.
23. Martinez A, Mosqueda-Taylor A, Marchesani FJ, 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:e25-9.
24. Carlos-Bregni R, Vargas PA, Santos Silva AR, Chaves-Netto HD, de Moraes M, Lopes MA. Adenomatoid odontogenic hamartoma: Concerns about correct nomenclature and 2 additional case reports. J Oral Maxillofac Surg 2009;67:1779-80.
25. Ali YH, Hussain AE. Adenomatoid odontogenic tumour of the middle turbinate: Case report and literature review. J Otolaryngol
Assessment of MRI and dynamic contrast-enhanced MRI in the differential diagnosis of adenomatoid odontogenic tumor. Eur J Radiol 2004;51:252-6.

30. McGuff HS, Alderson GL, Jones AC, Edgin WA. Oral and maxillofacial pathology case of the month. Adenomatoid odontogenic tumor. Tex Dent J 2008;125:1192-5.

31. Santos JN, Lima FO, Romério P, Souza VF. Adenomatoid odontogenic tumor: An unusual case exhibiting cribriform aspect. Quintessence Int 2008;39:777-81.

32. Ide F, Mishima K, Saito I, Kusama K. Rare peripheral odontogenic tumors: Report of 5 cases and comprehensive review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2008;106:e22-8.

33. Vasconcelos BC, Frota R, Cardoso AB, Porto GG, Carneiro SC. Adenomatoid odontogenic tumor. Braz J Otorhinolaryngol 2008;74:315.

34. Jivan V, Altini M, Meer S, Mahomed F. Adenomatoid odontogenic tumor (AOT) originating in a unicystic ameloblastoma: A case report. Head Neck Pathol 2007;1:146-9.

35. Chuan-Xiang Z, Yan G. Adenomatoid odontogenic tumor: Correlation of MRI with immunohistochemical study. Med Oral Patol Oral Cir Bucal 2006;11:E305-8.

36. Jham BC, Passos JB, Vieira do Carmo MA, Gomes CO, Mesquita RA. Adenomatoid odontogenic tumor originated in the periodontal ligament. Oral Oncol Extra 2006;42:268-71.

37. Nigan S, Gupta SK, Chaturvedi KU. Adenomatoid odontogenic tumor – A rare cause of jaw swelling. Braz Dent J 2005;16:251-3.

38. Matomehi MH, Shafeie HA, Azizi T. Salvage of an impacted canine associated with an adenomatoid odontogenic tumor: A case report. Br Dent J 2005;199:89-90.

39. Effiom OA, Odukoya O. Adenomatoid odontogenic tumour: A clinicopathological analysis and melanin pigmentation study of 31 Nigerian cases. Niger Postgrad Med J 2005;12:131-5.

40. Bravo M, White D, Miles L, Cotton R. Adenomatoid odontogenic tumor mimicking a dentigerous cyst. Int J Pediatr Otorhinolaryngol 2005;69:1685-8.

41. 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 tumour: A multicentric study. Oral Oncol 2005;41:835-42.

42. Sato D, Matsuoka K, Yama M, Kakizawa T, Inoue T. Adenomatoid odontogenic tumor arising from the mandibular molar region: A case report and review of the literature. Bull Tokyo Dent Coll 2004;45:223-7.

43. Batra P, Prasad S, Parkash H. Adenomatoid odontogenic tumour: Review and case report. J Can Dent Assoc 2005;71:250-3.

44. Asaumi J, Yanagi Y, Konouchi H, Hisatomi M, Matsuaki H, Shigehara H, et al. Assessment of MRI and dynamic contrast-enhanced MRI in the differential diagnosis of adenomatoid odontogenic tumor. Eur J Radiol 2004;51:252-6.

45. Walker LM, Wood AJ, McDonald A, Carpenter W. Unerupted mandibular second primary molar with an unusual histopathological finding: A case report. J Dent Child (Chic) 2004;71:77-9.

46. Olqag V, Köseoglu BG, Kasapoglu C. Adenomatoid odontogenic tumor: A report of an unusual maxillary lesion. Quintessence Int 2003;34:686-8.

47. 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.

48. Konouchi H, Asaumi J, Yanagi Y, Hisatomi M, Kishi K. Adenomatoid odontogenic tumor: Correlation of MRI with histopathological findings. Eur J Radiol 2002;44:19-23.

49. 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.

50. Takahashi K, Yoshino T, Hashimoto S. Unusually large cystic adenomatoid odontogenic tumour of the maxilla: Case report. Int J Oral Maxillofac Surg 2001;30:173-5.

51. Bulut E, Tasar F, Akkoçaglu M, Ruçan S. An adenomatoid odontogenic tumor with unusual clinical features. J Oral Sci 2001;43:283-6.

52. Blumenthal NM, Mostofi R. Repair of an intrabony defect from an adenomatoid odontogenic tumor. J Periodontol 2000;71:1637-40.

53. Lee JK, Lee KB, Hwang BN. Adenomatoid odontogenic tumor: A case report. J Oral Maxillofac Surg 2000;58:1161-4.

54. Damm DD, Fantasia JE. Failure of eruption. Adenomatoid odontogenic tumor. Gen Dent 2000;48:650, 722.

55. Zlotogorski A, Buchner A, Kaffe I, Schwartz-Arad D. Radiological features of central haemangioma of the jaws. Dentomaxillofac Radiol 2005;34:292-6.

56. Mohanty N, Routray S, Swain N, Ingale Y. Adenomatoid odontogenic tumor: An unusual case exhibiting cribriform aspect. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2005;99:732-6.

57. Ide F, Mishima K, Saito I, Kusama K. Diagnostically challenging epithelial odontogenic tumors: A selective review of 7 jawbone lesions. Head Neck Pathol 2009;3:18-26.

58. McGuff HS, Alderson GL, Jones AC, Edgin WA. Oral and maxillofacial pathology case of the month. Adenomatoid odontogenic tumor. Tex Dent J 2008;125:1192-5.

59. Shephard M, Coleman H. Simultaneous adenomatoid and keratocystic odontogenic tumours in a patient with Gorlin-Goltz syndrome. Aust Dent J 2014;59:121-4.
63. Marrelli M, Pacifici A, Di Giorgio G, Cassetta M, Stefanelli LV, Gargari M, et al. Diagnosis and treatment of a rare case of adenomatoid odontogenic tumor in a young patient affected by attenuated familial adenomatosis polyposis (aFAP): Case report and 5 year follow-up. Eur Rev Med Pharmacol Sci 2014;18:265-9.

64. Yamazaki M, Maruyama S, Abe T, Babkair H, Fujita H, Takagi R, et al. Hybrid ameloblastoma and adenomatoid odontogenic tumor: Report of a case and review of hybrid variations in the literature. Oral Surg Oral Med Oral Pathol Oral Radiol 2014;118:e12-8.

65. Mohanty S, Gulati U, Mediratta A, Ghosh S. Unilocular radiolucencies of anterior mandible in young patients: A 10 year retrospective study. Natl J Maxillofac Surg 2013;4:66-72.

66. Gomez RS, Castro WH, Gomes CC, Loyola AM. Adenomatoid odontogenic tumor associated with odontoma: A case report and critical review of the literature. Head Face Med 2013;9:20.

67. Kurra S, Gunupati S, Prasad PR, Raju YS, Reddy BV. An adenomatoid odontogenic cyst (AOC) with an assorted histoarchitecture: A unique entity. J Clin Diag Res 2013;7:1223-5.

68. Bhatt R, Dave J, Nalawade TM, Mallikarjuna R. Adenomatoid odontogenic tumour in mandible in a 14-year-old boy. BMJ Case Rep 2013;2013. pii: bcr2013010287.

69. Lee SK, Kim YS. Current concepts and occurrence of epithelial odontogenic tumours: I. Ameloblastoma and adenomatoid odontogenic tumor. Korean J Pathol 2013;47:191-202.

70. Narayanan VS, Naidu G, Ragavendra R, Mhaske-Jedhe S, Haldar M. Adenomatoid odontogenic tumor of the mandible with unusual radiographic features: A case report. Imaging Sci Dent 2013;43:111-5.

71. Angiero F, Crippa R. Adenomatoid odontogenic tumor: A case report with immunohistological profile. Anticancer Res 2013;33:2673-7.

72. Li BB, Xie XY, Jia SN. Adenomatoid odontogenic tumor with fibro-osseous reaction in the surrounding tissue. J Craniofac Surg 2013;24:e100-1.

73. Agarwal A, Giri KY, Alam S. The interrelationship of adenomatoid odontogenic tumour and dentigerous cyst: A report of a rare case and review of the literature. Case Rep Pathol 2012;2012:358609.

74. Harne JC, Pedeutour F, Raybaud H, Ambrosetti D, Fabas T, Lombardi T. Immunohistochemical features in adenomatoid odontogenic tumour: Review of the literature and first expression and mutational analysis of β-catenin in this unusual lesion of the jaws. J Oral Maxillofac Surg 2013;71:706-13.

75. Lee JS, Yoon SJ, Kang BC, Kim OJ, Kim YH. Adenomatoid odontogenic tumor associated with unerupted first primary molar. Pediatr Dent 2012;34:493-5.

76. Singh V, Goyal S, Sheikh S, Shambulingappa P, Singh B, Singh R. Adenomatoid odontogenic tumor with dentigerous cyst: Report of a rare case with review of literature. Contemp Clin Dent 2012;3 Suppl 2:S244-7.

77. Prakash AR, Reddy PS, Rajanikanth, Bavle RM. Concomitant occurrence of cemento-ossifying fibroma and adenomatoid odontogenic tumor with bilateral impacted permanent canines in the mandible. Indian J Dent Res 2012;23:434-5.

78. Vasudevan K, Kumar S, Vijayasamundeeswari, Vigneswari S. Adenomatoid odontogenic tumor, an uncommon tumor. Contemp Clin Dent 2012;3:245-7.

79. de Matos FR, Nonaka CF, Pinto LP, de Souza LB, de Almeida Freitas R. Adenomatoid odontogenic tumor: Retrospective study of 15 cases with emphasis on histopathologic features. Head Neck Pathol 2012;6:430-7.

80. Damm DD. Localized gingival enlargement. Adenomatoid odontogenic tumor. Gen Dent 2012;60:355, 357.

81. Sharma N, Passi S, Kumar VV. Adenomatoid odontogenic tumor: As an unusual mandibular manifestation. Contemp Clin Dent 2012;3 Suppl 1:S29-32.

82. Mutalik VS, Shreshthha A, Mutalik SS, Radhakrishnan R. Adenomatoid odontogenic tumor: A unique report with histological diversity. J Oral Maxillofac Pathol 2012;16:118-21.

83. Sekiya R, Yamazaki H, Izawa K, Kaneko A, Tsukinoki K. Case of adenomatoid odontogenic tumor during pregnancy. Tokai J Exp Clin Med 2011;36:124-7.

84. Anegundi RT, Radhika R, Patil S, Sahana BA. Adenomatoid odontogenic tumor: An uncommon location. Pediatr Dent 2011;33:437-9.

85. Bhullar RP, Brar RS, Sandhu SV, Bansal H, Bhandari R. Mandibular adenomatoid odontogenic tumor: A report of an unusual case. Contemp Clin Dent 2011;2:230-3.

86. John JB, John RR. Adenomatoid odontogenic tumor associated with dentigerous cyst in posterior maxilla: A case report and review of literature. J Oral Maxillofac Pathol 2010;14:59-62.

87. Soares EC, Costa FW, Neto IC, Bezzerra TP, do Socorro Vital Patrocínio RM, Alves AP. Rare hybrid odontogenic tumor in a 2-year-old child. J Craniofac Surg 2011;22:554-8.

88. Bhandari N, Kothari M. Adenomatoid odontogenic tumour mimicking a pericopal cyst in pregnant woman. Singapore Dent J 2010;31:26-9.

89. Schirmer I, Reichhart PA. Adenomatoid odontogenic tumor (AOT) of the mandible: A surgical follow-up. Mund Kiefer Gesichtschir 2007;11:291-4.

90. Farah-Klibi F, Ferichichi L, Beya Rassou H, Zairi I, Rameh S, Adouani A, et al. Adenomatoid odontogenic tumor: Two cases. Rev Stomatol Chir Maxillofac 2007;108:61-4.

91. Buch RS, Coerd W, Wahllman U. Adenomatoid odontogenic tumor in calcifying odontogenic cyst. Mund Kiefer Gesichtschir 2003;7:301-5.

92. 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.

93. Robledo J, Mazock JB. Oral and maxillofacial pathology case of the month. Adenomatoid odontogenic tumor. Tex Dent J 2011;128:308-9, 314-5.

94. Steensland HS. Epithelioma adamantium. J Exp Clin Med 1905;6:377-89.

95. Thoma KH. Tumors of odontogenic origin. In: Oral Pathology. St. Louis (MO): Mosby; 1941. p. 945-6.

96. James W, Forbes JG. An epithelial odontome. Proc R Soc Med 1909;2:166-75.

97. Harbitz F. On cystic tumors of the maxillae, and especially on radionade cystadenomas (ademantomas). Dent Cosm St. Louis (MO): Mosby; 1909;2:166-75.

98. Wohl MG. Tooth germ cysts of the jaw. Ann Surg 1916;64:672-9.

99. de Matos FR, Nonaka CF, Pinto LP, de Souza LB, de Almeida Freitas R. Adenomatoid odontogenic tumor: Retrospective study of 15 cases with emphasis on histopathologic features. Head Neck Pathol 2012;6:430-7.
