

# Mandibular adenomatoid odontogenic tumor: Radiographic and pathologic correlation

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## Abstract

Adenomatoid odontogenic tumor (AOT) is a rare tumor of epithelial origin comprising 3% of all the odontogenic tumors. It is a benign, painless, noninvasive, and slow-growing lesion, with a relative frequency of 2.2-13% and often misdiagnosed as an odontogenic cyst on clinical examination. AOT affects young individuals with a female predominance, occurs mainly in the second decade, and usually surrounds the crown of unerupted teeth. This lesion is most commonly located in the anterior maxilla and rarely in the mandible. It is usually associated with an impacted canine. AOT frequently resembles lesions like dentigerous cyst or ameloblastoma. AOT has three variants, follicular, extrafollicular, and peripheral. The intraoral periapical radiograph is the best radiograph to show radiopacities in AOT as discrete foci having a flocculent pattern within radiolucency even with minimal calcified deposits. These calcified deposits are seen in approximately 78% of the lesions. Herewith, we present the report of four unusual cases of AOT located in the mandible, with an emphasis on radiographic findings and on pathologic correlation, and on reviewing the existing literature on this tumor.

**Key words:** Adenomatoid odontogenic tumor, benign neoplasm, impacted tooth, mixed lesion, radiopaque foci

## INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a rare tumor of epithelial origin comprising 3% of all the odontogenic tumors.<sup>[1,2]</sup> It was first described by Steensland in 1905. In 1907, AOT was described as pseudo-adenameloblastoma by Dreibradt.<sup>[1]</sup> Stafne in 1948 considered AOT as a distinct entity, whereas others believed it to be a variant of ameloblastoma.<sup>[3,4]</sup> In 1969, Philipsen and Birn declined this thought and suggested the name 'adenomatoid odontogenic tumor'. In 1971, the World Health Organization (WHO) adopted the term 'adenomatoid odontogenic tumor'. Max and Stern, in 2003, coined the name 'adenomatoid odontogenic cyst'.<sup>[5]</sup> Various terms like adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum, and teratomatous odontoma were used before the term AOT.<sup>[2,3]</sup>

AOT is a benign, painless, noninvasive, and slow-growing tumor that does not infiltrate the bone.<sup>[5]</sup> Clinically, it is often misdiagnosed as an odontogenic cyst. The tumor appears as an intraoral-extraoral swelling in the maxilla and is sometimes referred to as 'two-third tumor' because it occurs in the maxilla in about two-third cases, about

two-third cases arise in young females, two-third cases are associated with an unerupted tooth, and two-third affected teeth are canines.<sup>[5,6]</sup> The origin of the AOT is controversial. It is thought to arise from odontogenic epithelium because it occurs in the tooth-bearing areas of the jaws, is often associated with the impacted tooth, and has various components of the enamel organ, dental lamina, reduced enamel epithelium, and/or their remnants.<sup>[4]</sup>

The purpose of this article is to report and analyze four unusual cases of AOT located in the mandible, with an emphasis on radiographic findings and with pathologic correlation, and to review the existing literature on this tumor.

## DISCUSSION

AOT occurs mainly in the second decade of life, and is uncommon in patients older than 30 years of age. Females are more commonly affected than males with a ratio of 2:1, but it was not so in our analysis.<sup>[3,7]</sup> This female predilection is even more marked in Asian populations, the highest female

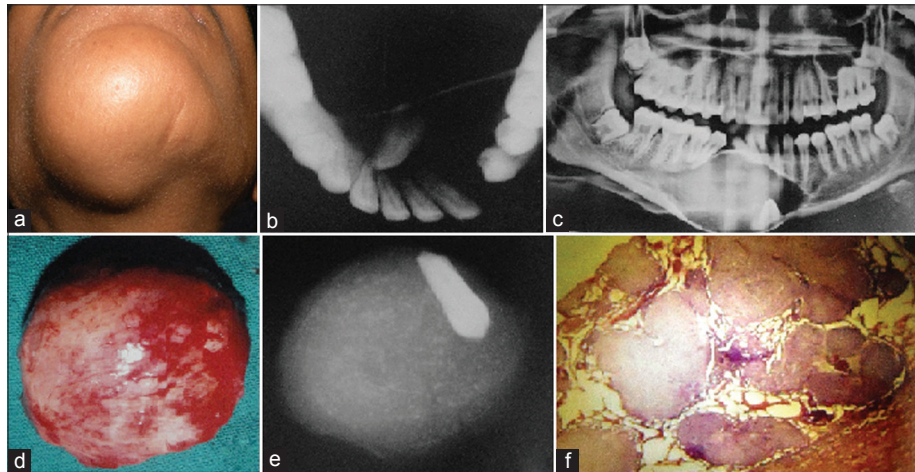
incidence being observed in Sri Lanka (3.2:1) and Japan (3:1). The maxillary arch is the predominant site of occurrence, being almost twice as frequent as that of the mandible, and the anterior part of the jaw is more frequently involved than the posterior part.<sup>[5]</sup> Giansanti *et al.* (1970) reported that 65% AOTs were seen in the maxilla and 35% in the mandible. Of

the maxillary lesions, 80% occurred in the anterior region, 14% in the premolar region, and few in the molar area. Of the mandibular lesions, 69% were found in the anterior region, 27% in the premolar region, and a few in the molar region.<sup>[8]</sup> It is pertinent to note that all our four cases had AOT in the anterior as well as posterior part of the mandible.

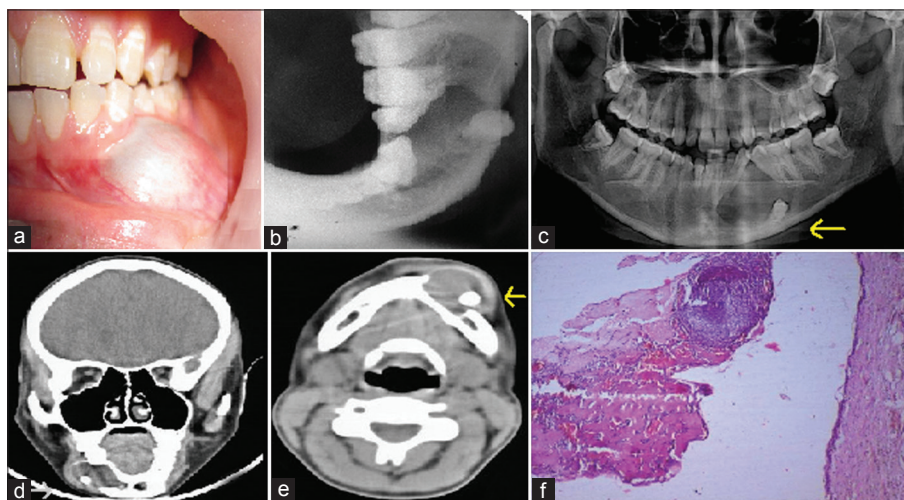
### CASE SYNOPSIS

Age (yrs.)	Gender	Jaw	Affected sites	Intraoral manifestation	Radiographic observations
14	Male	Mandible	Symphysis and parasymphysis [Figure 1a]	Nontender bony hard swelling from 32 to 44, obliterated labial vestibule due to expansion of the buccal cortical plate, and missing 43 were noted.	<p>The mandibular true occlusal [Figure 1b] and panoramic radiograph [Figure 1c] revealed an extensive well-defined circumscribed, corticated unilocular expansile radiolucent lesion extending from 34 to 46 with an impacted 43, which is displaced linguo-inferiorly toward the lower border of the mandible. The adjoining teeth show inclination. The expansion of lingual cortical plate is noted with no discontinuity. The lesion presented with multiple radiopaque flecks of calcified material.</p> <p>The surgically excised mass [Figure 1d] was subjected to radiographic image which showed radiopaque multiple flecks of calcified material and impacted 43 [Figure 1e].</p>
18	Female	Mandible	Symphysis and parasymphysis	Nontender bony hard buccal swelling from 33 to 36, obliterated buccal vestibule, and missing 34 were noted [Figure 2a].	<p>The mandibular true occlusal [Figure 2b] and panoramic radiograph [Figure 2c] revealed a well-defined circumscribed, corticated unilocular expansile radiolucent lesion extending from 31 to 36 with an impacted 34 which is displaced bucco-inferiorly toward the lower border of the mandible. The adjoining teeth show inclination. The expansion of buccal cortical plate is noted with no discontinuity. The lesion has radiopaque flecks of calcified material.</p> <p>The plain computed tomogram (axial and coronal) shows a hypodense area in the left parasymphysis and body of the mandible. The buccal cortical plate is thinned, expanded, and continuous. Within the lesion, a single hyperdense mass is noted, suggestive of an impacted tooth. [Figure 2d and e]</p>
32	Male	Mandible	Left parasymphysis	Nontender bony hard buccal swelling from 35 to 44, obliterated buccal vestibule due to expansion of the buccal cortical plate, and missing 31, 32, 33, 41, 42, 43 were noted [Figure 3a].	<p>The mandibular true occlusal [Figure 3b] and panoramic radiograph [Figure 3c] revealed an extensive well-defined, circumscribed, homogeneous, corticated unilocular radiolucent expansile lesion extending from 36 to 44 with impacted 31, 32, 33, 41, 42, 43 which is displaced linguo-inferiorly toward the lower border of the mandible. The adjoining teeth show inclination. The expansion of buccal and lingual cortical plate was noted with no discontinuity.</p> <p>Computed tomography (axial and coronal) revealed an osteolytic lesion involving the anterior mandible with intact cortical borders. The margins were thin and continuous with no perforation. Multiple teeth were seen within the mass. [Figures 3d and e]</p>
28	Male	Mandible	Body	Tender hard buccal swelling from 44 to 47, obliterated buccal vestibule, and missing 45 were noted. [Figure 4a]	<p>The mandibular true occlusal [Figure 4b] and panoramic radiograph [Figure 4c] revealed an extensive well-defined corticated unilocular expansile radiolucent lesion extending from 43 to 47 with impacted 45 which is displaced buccally. The expansion of buccal and lingual cortical plate was noted with no discontinuity. The adjoining teeth show inclination, and root resorption was evident in 44, 46, and 47. The inferior border of the mandible was intact.</p> <p>Computed tomography (coronal and axial) revealed a unilocular osteolytic lesion with perforated buccal and lingual cortical plates. A tooth was visible along the lower margin of the lesion. [Figure 4d and e]</p>

(Note: FDI teeth numbering system has been used)



**Figure 1:** (a) The extraoral photograph shows a well-defined hard swelling present on chin and submental region (b) The mandibular occlusal radiograph shows lingual cortical plate expansion and an impacted 43 (c) Panoramic radiograph shows well-defined corticated expansile lesion, thinning of inferior border, and impacted 43 (d) Surgically excised mass (e) Radiographic image of the excised mass showing small multiple flecks of calcification and impacted 43 (f) Photomicrograph (H and E,  $\times 40$ ) showing proliferation of columnar and cuboidal cells in whorls



**Figure 2:** (a) Intraoral photograph showing well-demarcated swelling in the buccal vestibule (b) Mandibular occlusal radiograph shows buccal cortical plate expansion with impacted 34 (c) Panoramic radiograph shows well-defined corticated expansile lesion with impacted 34 (d and e) Computed tomogram (coronal and axial) shows a hypodense lesion with expansion of buccal cortical plate (f) Photomicrograph (H and E,  $\times 20$ ) showing a whorl of columnar and cuboidal cells, thin nonkeratinized epithelium and cyst-like spaces

AOT is frequently associated with an impacted tooth, a canine in more than 60% of the cases.<sup>[4]</sup> Permanent incisors, premolars, molars, and deciduous teeth are rarely involved. But more than one tooth may also be related with AOT as noticed in our case analysis wherein permanent incisors, canines, and premolars were involved with the lesion.

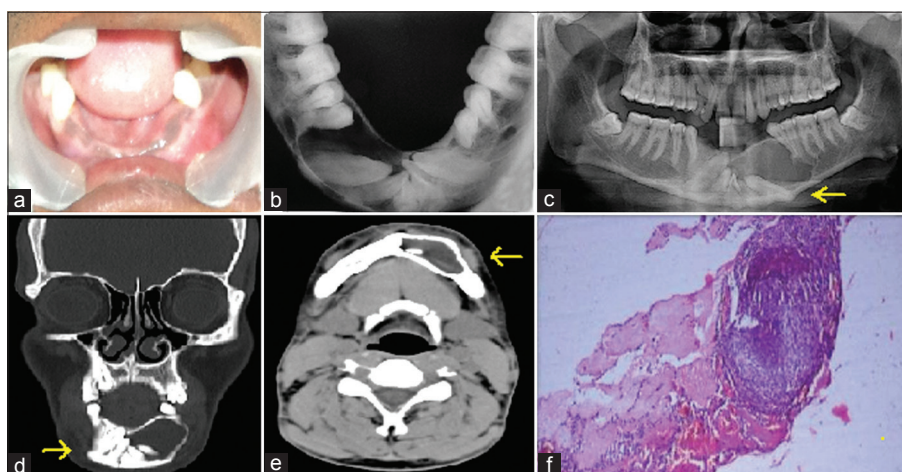
AOTs are relatively small in size. Usually, they do not exceed 1-3 cm in diameter.<sup>[9]</sup> However, some large tumors have been reported, and all our present cases had unusually large dimensions, that is, more than 3 cm.

The continuous slow growth of the lesion may cause cortical plate expansion leading to a painless hard swelling,

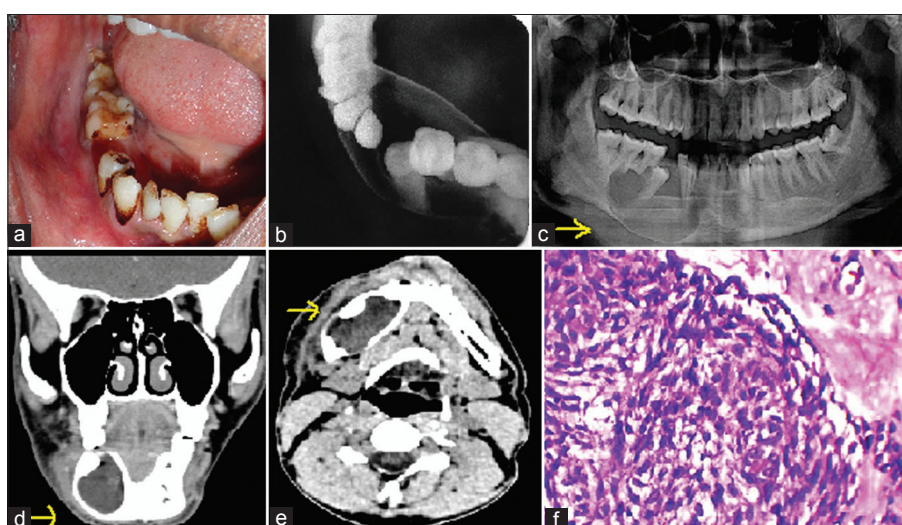
asymmetry of the face, and displacement of the teeth, as was evident in our case analysis. As the growth is only within the confinement of the jaw bone, there is no invasion in the soft tissue. The slow-growing nature of the lesion may cause the patients to tolerate the swelling for years until it produces an obvious deformity. Delayed eruption of a permanent tooth or a regional swelling of the jaws may be the first symptom. Pain or other neurologic signs are not characteristic. Clinically, Ajagbe *et al.* (1985) found that a few lesions on palpation were soft and spongy like cysts, whereas many lesions were firm and bony hard, like fibro-osseous lesions.<sup>[10]</sup>

Generally, AOT occurs intraosseously, but can also occur





**Figure 3:** (a) Intraoral photograph showing edentulous space in anterior region (b) Mandibular occlusal radiograph shows a radiolucent lesion with six impacted teeth and buccal-lingual cortical plate expansion (c) Panoramic radiograph shows well-defined corticated expansile lesion with multiple impacted teeth (d and e) Computed tomogram (coronal and axial) shows a well-defined osteolytic lesion with impacted teeth (f) Photomicrograph (H and E,  $\times 20$ ) showing a whorl of columnar cells and cuboidal cells and fibrous connective tissue



**Figure 4:** (a) Intraoral photograph showing missing 45 and an apparent vestibular obliteration (b) Mandibular occlusal radiograph showing a radiolucent lesion with buccal-lingual cortical plate expansion and impacted 45 (c) Panoramic view shows a well-corticated lesion with an impacted 45 (d and e) Computed tomogram (coronal and axial) shows a well-defined osteolytic lesion, impacted teeth, and ruptured lingual cortical plate (f) Photomicrograph (H and E,  $\times 40$ ) showing a whorl of columnar cells and cuboidal cells, nonkeratinized epithelium, and fibrous connective tissue

rarely in peripheral locations. There are three variants of AOT: Follicular, extrafollicular, and peripheral. The follicular type (pericoronal) is a central intrabony lesion associated with an unerupted tooth, which accounts for about 70% of all cases. The extrafollicular type (extracoronal) is also an intraosseous lesion, but unrelated to an unerupted tooth, and represents 25% of all AOTs. The peripheral type (extra osseous) is a rare form that arises in the gingival tissue, and accounts for 5% of all AOTs.<sup>[3,5,6]</sup> All the cases involved in the present analysis were of the follicular variety.

All the variants of AOT show identical histological features.<sup>[10]</sup> The histological typing of WHO defined AOT as a tumor of odontogenic epithelium with duct-like

structures and with varying degrees of inductive change in the connective tissue.<sup>[1]</sup> AOT is usually surrounded by a well-developed connective tissue capsule. It may present as a solid mass, a single large cystic space, or as numerous small cystic spaces.<sup>[1]</sup> The tumor is composed of spindle-shaped or polygonal cells forming sheets and whorled masses in a scant connective tissue stroma. The amorphous eosinophilic material is seen between the epithelial cells, as well as in the center of the rosette-like structure. The characteristic duct-like structures are lined by a single row of columnar epithelial cells, the nuclei of which are polarized away from the central lumen, as was evident in all our cases. The lumen may be empty or contain amorphous eosinophilic material.<sup>[3,5]</sup> Dystrophic calcification in varying amounts and

in different forms is usually encountered in most AOTs within the lumina of the duct-like structures, scattered among epithelial masses or in the stroma. [Figures 1f, 2f, 3f and 4f] The immunohistochemical studies report that the slow growth, benign character, and low tendency to recur are clearly related to the low cellular proliferation observed on performing immunostaining for the Ki67 antigen.<sup>[10]</sup>

Radiographically, the intraosseous AOT has distinct features. It usually appears as a pericoronal well-circumscribed unilocular radiolucency or radiopaque-radiolucent mixed lesion with well-defined corticated or sclerotic border, usually surrounding an unerupted tooth, and may contain multiple minute variable-shaped calcifications or radiopaque foci, which may appear like a 'cluster of small pebbles'. These calcified deposits are seen in approximately 78% of the lesions.<sup>[7,9]</sup> Rarely the lesion manifests with no radiopaque component, as seen in two of our cases.

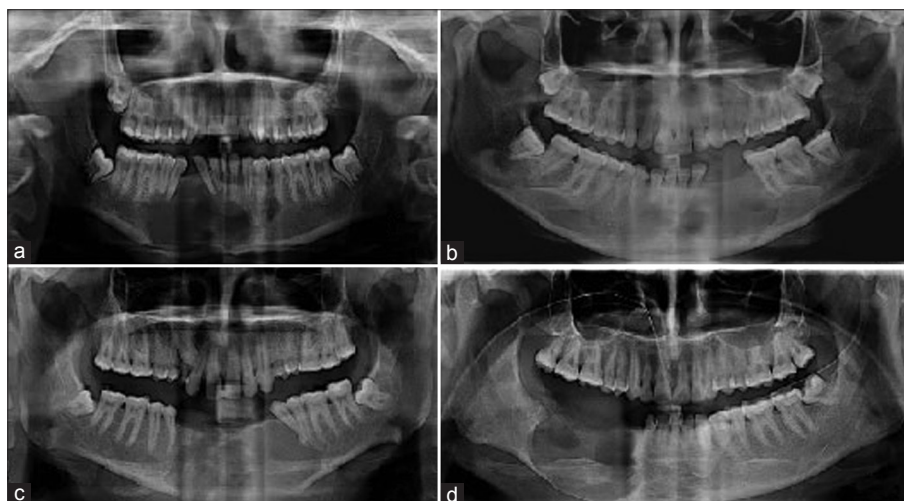
Small calcifications within the tumor are not seen on radiographs; so the lesion is completely radiolucent and mimics a dentigerous cyst in growth pattern and appearance. However, an AOT often appears to envelop the crown as well as the root, unlike the dentigerous cyst which does not envelop roots.<sup>[3]</sup> Irregular root resorption is rarely seen, but two cases in the present analysis showed this feature distinctly.<sup>[4,5]</sup> The extraosseous AOTs are rarely detected radiographically, but slight erosion of the underlying alveolar bone cortex maybe seen.<sup>[5]</sup> Comparing the diagnostic accuracy, Dare *et al.* found that the intraoral periapical radiograph is the best radiograph to show radiopacities in AOT as discrete foci having a flocculent pattern within radiolucency even with minimal calcified deposits, when compared to a panoramic radiograph.<sup>[4,7,8]</sup> In addition, magnetic resonance imaging (MRI) is useful to distinguish AOT from other lesions.

The radiographic findings of AOT frequently resemble lesions such as dentigerous cyst, calcifying odontogenic cyst, calcifying epithelial odontogenic tumor, globulomaxillary cyst, unilocular ameloblastoma, ameloblastic fibro-odontoma, odontogenic keratocyst, and intermediate-stage odontoma.<sup>[6]</sup>

The surgical management of this tumor should be enucleation along with the associated impacted tooth and simple curettage.<sup>[7]</sup> Conservative treatment is adequate because the tumor is not locally invasive, is well encapsulated, and is separated easily from the bone. The surgical specimen may be solid or cystic. The recurrence rate is as low as 0.2%.<sup>[8-10]</sup> However, in exceptional cases of large tumors or risk of bone fracture, partial resection, en bloc of the mandible or maxilla has been indicated. In addition, the use of lyophilized bone and guided tissue regeneration are recommended in large osseous cavities. The prognosis is excellent in majority of the cases. The cases described here have been on regular follow-up since 12-24 months after surgery and no recurrence is noted [Figure 5].

## CONCLUSION

AOT is a rare slow-growing painless, noninvasive tumor, often misdiagnosed as an odontogenic cyst. Although it affects young individuals, mainly females, commonly found in the anterior maxilla and associated with an impacted canine, this was not so in our analysis. Interestingly, our present cases had some unusual clinical and radiographic features that distinguished it from most normal types of AOT. The intraoral periapical radiograph was the best radiograph to show radiopacities in AOT as discrete foci having a flocculent pattern within radiolucency even with minimal calcified deposits. It



**Figure 5:** (a-d) Panoramic view showing normal healing process

should be emphasized that although AOT is very rare, careful diagnosis and adequate interpretation of clinical and radiographic findings may be helpful in arriving at a correct diagnosis.

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