

Aggressive adenomatoid odontogenic tumor of the mandible: A rare case report and review of the literature

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Abstract

Adenomatoid odontogenic tumor (AOT) is a relatively rare and distinct odontogenic tumor that is exclusively odontogenic epithelium in origin. Although considerable numbers of reports are available with regard to the clinical and histological spectrum of AOT, very few have highlighted its rare aggressive nature. This article focuses on an AOT causing jaw swelling in the mandibular anterior region with cortical plate expansion and perforation, root resorption and paresthesia. The present case had remarkably unusual clinical and radiographic features that distinguished it from most conventional AOTs and supported its neoplastic nature.

Keywords: Adenomatoid odontogenic tumor, aggressive, mandible

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INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a relatively rare, distinctly benign odontogenic neoplasm that comprises only 0.1% of tumors of head and neck and 3%–7% of all odontogenic tumors.^[1] In 1969, Philipsen and Birn introduced the term AOT which was adopted by the WHO classification in 1971. Topographically, the AOT occurs in peripheral and central variants, the latter further in follicular and extrafollicular types. It is sometimes referred to as “two-thirds tumor” because it occurs in the maxilla in about 2/3 cases, about 2/3 cases arise in young females, 2/3 cases are associated with an unerupted tooth and 2/3 of the affected teeth are canines.^[2] The tumor appears as a well-circumscribed, painless swelling and its discovery is often fortuitous during routine radiological examinations. However, some large tumors have been reported.^[3] Here, we report a case of mandibular AOT which shows unusual,

aggressive behavior suggesting it to be a true benign but an aggressive neoplasm and a review of literature on aggressive AOTs.

CASE REPORT

A 24-year-old female patient reported to our department with a gradually increasing, painless swelling in the lower anterior jaw region for 1 month. There was paresthesia on the left side of lower lip. Extraoral examination showed a diffuse painless swelling in mandibular symphysis region giving the patient's profile a prognathic appearance. On palpation, the swelling was bony hard in consistency, nonfluctuant, nontender and showed no evidence of discharge on digital pressure. There was no regional lymphadenopathy. The patient's social, family and habit histories were noncontributory. Intraoral examination revealed a bony hard swelling extending from the

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permanent right mandibular canine to the left mandibular second premolar causing buccal expansion with obliteration of the buccal vestibule. The permanent mandibular right central incisor was missing. Overlying mucosa appeared blanched with Grade I mobility of the permanent left central and lateral incisors. No intraoral draining sinus was noted [Figure 1].

A panoramic radiograph demonstrated a well-demarcated unilocular radiolucent lesion measuring about 6.5 cm × 3.5 cm in size extending from the permanent right first premolar to the left second premolar. There was displacement of mandibular anterior teeth and external root resorption of the right lateral incisor, canine and left central and lateral incisor. The impacted permanent right central incisor was displaced as far as the inferior border of the mandible. Thinning and expansion of the inferior mandibular cortex was seen [Figure 2]. Cone-beam computed tomography showed a well-defined, unilocular, corticated osteolytic lesion with bicortical expansion and buccal cortical plate perforation. No radiographically detectable calcifications were seen in the lesion [Figure 3].

The massive size of this lesion attained within a short period with cortical perforation and associated paresthesia, suggested an aggressive intraosseous neoplasm.

Based on the clinical and radiographic findings, differential diagnosis of unicystic ameloblastoma, keratocystic odontogenic tumor, dentigerous cyst, central giant cell lesion and odontogenic myxoma was considered.

Macroscopic and microscopic features

Incisional biopsy was performed under local anesthesia. Hematoxylin and eosin-stained, formalin-fixed, paraffin-embedded sections under microscope revealed tumor mass composed of spindle-shaped cells forming sheets and whorled masses, solid nodules of cuboidal epithelial cells forming rosette-like structures in a scant connective tissue stroma. Also, present were duct-like structures lined by a single row of columnar epithelial cells with the nuclei polarized away from the luminal surface. Interspersed between the epithelial cells was amorphous eosinophilic material [Figure 4]. Based on these distinctive histomorphological features, a diagnosis of AOT was made.

Enucleation under general anesthesia was planned. The tumor was removed along with impacted central incisor. Teeth associated with the lesion and showing resorbed roots were also extracted. Grossly, the surgical specimen was tan colored, roughly spherical in shape measuring 6 cm × 3 cm in diameter. Cut surface revealed a cystic



Figure 1: Diffuse swelling causing obliteration of labial vestibule with missing permanent mandibular right central incisor

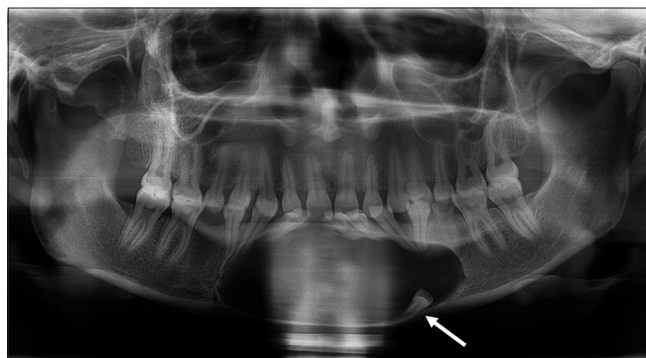


Figure 2: Panoramic radiograph showing impacted permanent mandibular right central incisor in a well-defined radiolucency, expansion and thinning of the inferior border of the mandible, displacement and resorption of the anterior teeth

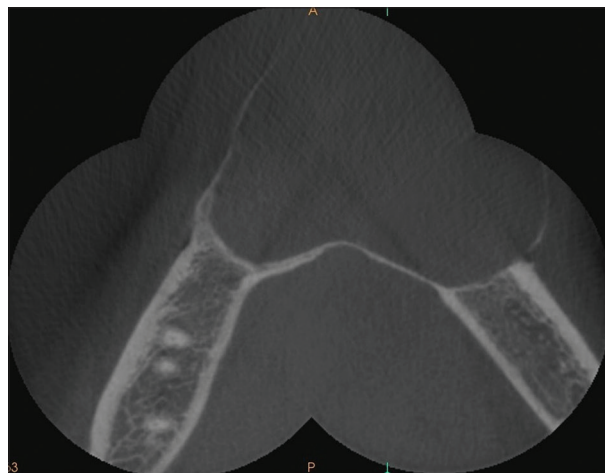


Figure 3: Cone-beam computerized tomography reveals buccolingual expansion, thinning and erosion of buccal cortical plate

space with the permanent mandibular right central incisor embedded in it [Figure 5]. Cystic wall was thick measuring 9 mm with hemorrhagic material in the lumen. The histopathological features were in conformation with that

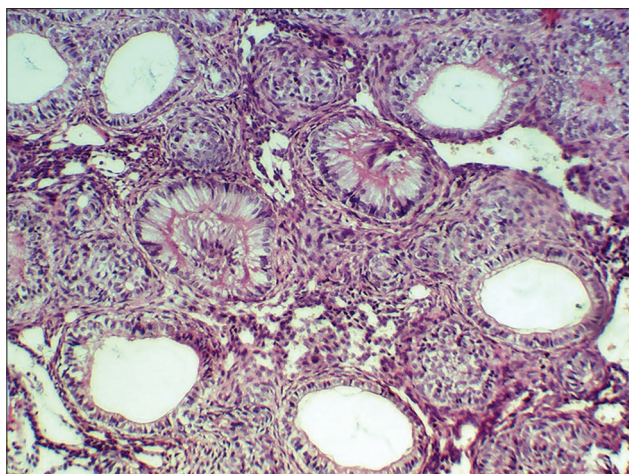


Figure 4: Photomicrograph illustrates a highly cellular tumor mass with multinodular proliferation of spindle, cuboidal, columnar and stellate reticulum-like cells forming nests of rosette-like and duct-like structures (H and E stain, x10)

of incisional biopsy and the final diagnosis of follicular variant of AOT was established. Postoperative healing was uneventful. The patient was subsequently lost to follow-up.

DISCUSSION

In the 2005 WHO classification of odontogenic tumors, AOT was classified into the first group of tumors (odontogenic epithelium without ectomesenchyme). Because of the absence of ectomesenchyme in immunohistochemical staining and dysplastic dentin, AOT is now considered the result of metaplastic process rather than epithelial-ectomesenchyme interaction.^[4]

The histogenesis of AOT is still uncertain; there has long been a debate as to whether it represents anomalous hamartomatous growth or a true benign neoplasm.^[5,6]

The present case had some unusual clinical and radiographic features that distinguished it from most conventional AOTs and supported its neoplastic nature.

An intensive literature search revealed very few reports where this tumor had such a locally destructive presentation.

This case was seen in a female in the 2nd decade of life that coincides well with the literature. It was present in the mandible that, although not rare, is an unusual site of presentation for an AOT. Furthermore, it did not follow the trend of an indolent benign tumor. There was marked displacement with substantial mobility and root resorption of the teeth in contact with the tumor mass. AOT generally does not exceed 1–3 cm in diameter.^[2] The most interesting finding in our case was the rapid progression to more than 6 cm within 1 month with associated unilateral paresthesia

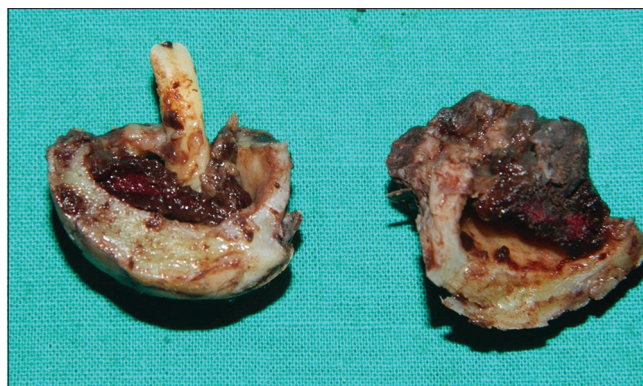


Figure 5: Gross appearance revealing a tan colored, roughly spherical-shaped tumor mass with embedded root of mandibular right central incisor

indicating compression of the mental nerve. The buccal cortex showed considerable expansion and perforation in several places. Thinning and expansion of inferior border of the mandible was also noted. The impacted tooth associated with the lesion was mandibular central incisor. Literature analysis revealed canines to be the most common teeth associated with the follicular AOT, with maxillary canines accounting for 51.3% of all cases, followed by incisors, premolars and molars.^[3] Although the aggressive behavior of the reported lesion is quite exceptional, the histopathology is identical to that of conventional AOT.

Conservative surgical enucleation or curettage is the treatment of choice for AOT. In the present case in spite of the locally destructive presentation, enucleation was planned bearing in mind the patients' age and psychological well-being coupled with rare recurrence and absence of malignant potential of this lesion.

Clinically such an aggressive variant of AOT is very rare. To the best of our knowledge, only 7 cases in the mandible^[3,7-12] and 3 in the maxilla^[13-15] have been reported showing aggressive AOTs. The clinical and radiological characteristics of these tumors are listed in Table 1. The final diagnosis of an AOT was arrived only after histopathological intervention. What is intriguing is that the histology has always remained identical with remarkable consistency irrespective of the biological behavior of the tumor. Due to this distinctive histomorphology, the diagnosis can always be made with ease.

CONCLUSION

Although uncommon, mandibular AOT is not rare. Consequently, in cases of aggressive unilocular radiolucent lesions in the mandibular anterior region, especially when associated with an impacted tooth, AOT may be considered as a differential diagnosis.

Table 1: Aggressive adenomatoid odontogenic tumors as reported in the literature

| Authors | Location | Course | Clinical features | Impacted tooth | Radiographic features | Root resorption | Treatment |
|---|----------|------------|---|----------------|--|------------------------------|---|
| Nomura <i>et al.</i> ^[3] | Mandible | 2 years | Vascular appearing granulomatous tumor. Size: 5 cm × 3.5 cm. Extent: 43 to right ramus. Hypoesthesia. Displacement of teeth from 33 to 41 | - | Well-defined radiolucent lesion. Inferiorly displaced inferior alveolar canal, buccal cortical plate destruction | 33 | <i>En bloc</i> resection |
| Geist and Mallon ^[7] | Mandible | 10 days | Bony hard, nontender swelling obliterating labial vestibule. Extent: 35 to 45. Displacement of 31, 41. Retained 83 | 43 | Well-defined radiolucent lesion with small irregular radio-opaque foci. Expansion and thinning of inferior cortex, perforation of buccal cortex | 33-43 | Enucleation |
| Khot and Vibhakar ^[8] | Mandible | 2 years | Bony hard, nontender swelling obliterating buccal and lingual vestibule. Size: 7 cm × 15 cm. Extent: 32-47. Pus discharge. Retained 83 | 43 | Well-defined unilocular radiolucent lesion. Expansion of cortical plates | 32-47 | Surgical curettage |
| Saluja <i>et al.</i> ^[9] | Mandible | 1 year | Bony hard, nontender swelling obliterating buccal vestibule. Size: 2 cm × 3 cm. Extent: 41-46. Mobility 42, 43, 44, 45, 46. Displacement: 43, 44 | - | Large well-defined radiolucent lesion. Buccal cortical plate expansion | 43, 44, 45, 46 | Enucleation |
| Shah <i>et al.</i> ^[10] | Mandible | 2 months | Bony hard, nontender swelling obliterating buccal and lingual vestibule. Size: 2.6 cm × 2 cm. Extent: 42-46. Displacement of 43, 44 | - | Well-defined radiolucent lesion. Bicortical expansion and thinning | 44, 45 and mesial root of 46 | Enucleation |
| Narayan <i>et al.</i> ^[11] | Mandible | 5-6 months | Bony hard, nontender swelling obliterating buccal and lingual vestibule. Size: 7 cm × 5 cm. Extent: 36-46. Displacement from 34 to 44. Grade II mobile 41, 42, 32. Grade I mobile 36, 31 | 31 | Well-defined radiolucent lesion with multiple, scattered radio-opaque foci and dense septae in lingual cortex giving a multilocular appearance. Bicortical expansion and thinning. Buccal plate perforation. Thinning of inferior cortex | 34, 35, 36, 44, 45, 46 | Enucleation |
| Qari <i>et al.</i> ^[12] | Mandible | 5 months | Bony hard, nontender swelling obliterating buccal and lingual vestibule. Size: 7 cm × 6 cm × 4 cm. Extent: 36-46. Class III mobility from 35 to 45. Displacement from 33 to 43. Paresthesia | 33 | Well-defined radiolucent lesion with dense radio-opacities. Thinning, expansion, scalloping of inferior mandibular cortex, perforation of buccal cortex | 34-45 | Enucleation |
| Takigami <i>et al.</i> ^[13] | Maxilla | 3 months | Diffuse swelling in left maxilla extending into maxillary sinus. Left nasal obstruction and visual disturbance. Left anosmia | - | Low-density mass filling up the left maxillary, left posterior ethmoid and sphenoid sinuses; extending into the left middle fossa with bony destructions and upward extension of pituitary complex | - | Subtotal removal of the tumor and decompression of the optic canal and superior orbital fissure |
| Takahashi <i>et al.</i> ^[14] | Maxilla | 10 months | Left nasal obstruction. Size: 6 cm × 6 cm × 5 cm. Extent: Left maxillary sinus and left nasal cavity | 28 | Large radiolucent lesion. Thinning and perforation of left bony sinus wall | - | Marsupialization followed by enucleation |
| Shetty <i>et al.</i> ^[15] | Maxilla | Accidental | Swelling causing obliteration of buccal vestibule. Extent: 11-15. Displacement of 15 | 13, 14 | Unilocular radiolucent lesion. Bicortical expansion, involving entire right maxillary sinus | 12 | Enucleation |
| Present case | Mandible | 1 month | Bony hard, nontender swelling obliterating buccal vestibule. Size: 6.5 cm × 3.5 cm. Extent: 35-45. Displacement from 35 to 45. Grade I mobile 31, 32 | 41 | Well-defined radiolucent lesion. Bicortical expansion, buccal cortical plate perforation. Thinning and expansion of inferior border of mandible | 31, 32, 42, 43 | Enucleation |

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.

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Conflicts of interest

There are no conflicts of interest.

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