

An Unusual Presentation of “Two-Thirds Tumor” in the Mandible – A Diagnostic Quandary with a Review of the Literature

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Abstract

An adenomatoid odontogenic tumor (AOT) is an uncommon benign tumor of the oral cavity commonly found in the maxillary anterior region and is associated with impacted canines in young females. It rarely occurs in the mandibular region with no impacted or missing teeth. A 21-year-old female reported to the clinic with swelling on the right side of the face for the past six months with no history of pain. Radiographic features such as unilocular radiolucency with thinning of the cortical borders and considerable buccal cortical expansion, as well as some evidence of radiopaque specks were noted. Histopathological examination revealed cells with hyperchromatic nuclei, rosette-like structures with focal areas of calcified mass, and concentric rings resembling Liesegang rings, suggestive of AOT. The tumor was treated surgically by enucleation and cauterization. Although follicular type is a common variant, the tumor presented in this case was of extrafollicular type noted in the mandibular canine and premolar regions of a young female patient with no related impacted tooth.

Keywords

Adenomatoid Odontogenic Tumor; Extrafollicular; Mandible; Odontogenic Tumor

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Introduction

Oral tumors may be odontogenic or non-odontogenic in origin. The World Health Organization currently classifies adenomatoid odontogenic tumor (AOT), also known as adenoameloblastoma or ameloblastic adenomatoid tumor, as a benign odontogenic tumor made up of odontogenic epithelium with epithelial, mesenchymal, or ectomesenchymal components [1].

AOT, which accounts for between 2.2 and 7.1% of all odontogenic tumors, was first described by Philipsen and Birn in 1969. As it developed from the enamel organ, they classified it as an epithelial odontogenic tumor [2]. AOTs are thought to be rare, non-aggressive lesions with gradual, steady progression. These lesions present clinically as a painless swelling that progressively increases in size and affects the bone [3].

AOTs are more common in younger age groups, with two-thirds of cases identified in the second decade of life and more than half of cases diagnosed in the age range

of 13-19 years [4]. The female predilection of the tumor is noted, as more than two-thirds of cases are diagnosed among females (2:1) [5]. The maxillary anterior region is the most prevalent location for this tumor, accounting for 65% of all cases. The remaining 35% of cases, however, occur in the mandible [6]. It is strongly associated with an impacted or unerupted tooth and it rarely develops distal to the premolar [7].

Radiographically, these lesions appear as circumscribed, unilocular radiolucent lesions with radiopaque borders. Calcification inside the lesion, which gives the lesion a mixed radiographic appearance, helps distinguish AOT from other lesions [3]. Unlikely calcification specks are seen inside the radiolucency [8].

Histologically, they are composed of spindle-shaped epithelial cells arranged in a rosette-like pattern inside a fibrous stroma and encased in a fibrous capsule. Cells exhibiting reversal of polarity, such as cuboidal or columnar cells, are suggestive of secretory activity and make up the tumor core. Throughout the tumor, calcification foci

may be recognized [4]. Conservative treatment by enucleation or curettage is indicated [9, 10] since, according to the literature, the recurrence rate of these tumors is as low as 0.2% [11], with one out of 1,558 cases of AOTs showing recurrence, which signifies the fact that these tumors have a relatively lower recurrence rate as compared to other odontogenic tumors. We report a case of a rare extra-follicular variant of AOT in the mandibular anterior region of a 21-year-old female, with a review of the literature.

Case Report

A 21-year-old female patient presented to the Department of Oral Medicine and Radiology with a primary complaint of painless swelling on the right side of the face for the past six months. The swelling was initially small in size and progressively grew to its current size. The patient provided no substantial medical or personal history.

On extraoral examination, facial asymmetry with a diffuse swelling measuring approximately 3 x 2 cm was seen in the right parasymphysis region, extending from the right corner of the mouth to the lower border of the mandible (Fig. 1a). The swelling was firm in consistency, non-tender,

roughly oval, non-compressible, and fixed.

Intraoral examination revealed a dome-shaped swelling with buccolingual expansion from the gingival margin up to the vestibule, extending from tooth # 42 to tooth # 45 (Fig. 1b). The expansion was approximately 5 x 3 cm in size, with obliteration of the adjacent buccal vestibule. The overlying mucosa appeared to be normal in color and texture. The swelling was firm and non-tender on palpation. Miller’s Grade III mobility was associated with teeth #s 43, 44, and 45 being depressible in the socket. Displacement of teeth #s 43, 44, and 45 was observed, and the associated teeth were non-vital. The history and clinical presentation were suggestive of benign lesions. Ameloblastoma, odontogenic keratocyst, and adenomatoid odontogenic tumor were considered for clinical differential diagnosis [12].

X-ray examinations included an intraoral periapical radiograph (IOPA), a true occlusal radiograph, and an orthopantomogram. The IOPA revealed displacement of root fragments of teeth #s 43 and 45 due to the pressure exerted on both sides by the expansile lesion (Fig. 2a). The mandibular true occlusal radiograph revealed unilocular radiolucency with thinning of the cortical borders extending from the mesial side of teeth #s 43 to 45, with considerable buccal cortical expansion, mimicking an eggshell effect with some evidence of radiopaque specks lingually (Fig. 2b). Orthopantomogram revealed an osteolytic radiolucent lesion, delimited in the region of the right mandible, measuring about 5 x 3 cm, and extending from teeth #s 43 to 45. Loss of lamina dura was observed (Fig. 2c). As there was no associated impacted tooth, calcifying epithelial odontogenic cysts or tumors were considered in the radiographic diagnosis.



Figure 1. a) preoperative frontal facial view revealing swelling on the right side of the face; b) intraoral view depicting swelling on the right buccal vestibule.



Figure 2. a) Intraoral periapical radiograph revealing displacement of root fragments of teeth #s 43 and 44 by the bony lesion; b) Mandibular true occlusal view depicting eggshell effect due to thinning of the buccal cortex; c) orthopantomogram showing the tumor extending to 0.8 cm from the lower border of the mandible.



Figure 3. Intraoperative photographs taken during surgical enucleation (a, b); during bone graft placement (c); after suture placement (d); with the excised specimen in the center (e).

Conservative surgical enucleation along with chemical cauterization using Carnoy’s solution was performed under general anesthesia, as seen in Fig. 3a,b. Carnoy’s solution was used, as it is a non-selective fixative that affects both tumor cells and surrounding tissues by penetrating into the cancellous bone spaces, devitalizing and fixing remaining tumor cells, thereby preventing the need for a second surgery [13]. An enucleated mass measuring 2.7 x 2.6 cm² (Fig. 3e) and extracted tooth # 44 were radiographically examined and then sent for histopathological examination.

Histopathology revealed cuboidal to polygonal cells with hyperchromatic nuclei forming nests and rosette-like structures (Fig. 4a). There was noted a tubular or duct-like appearance with the spaces lined by a single row of tall columnar cells with polarity reversal (Fig. 4b). In the foci area, calcified masses with concentric rings, resembling Liesegang rings, were visible within the tumor islands (Fig. 4c).

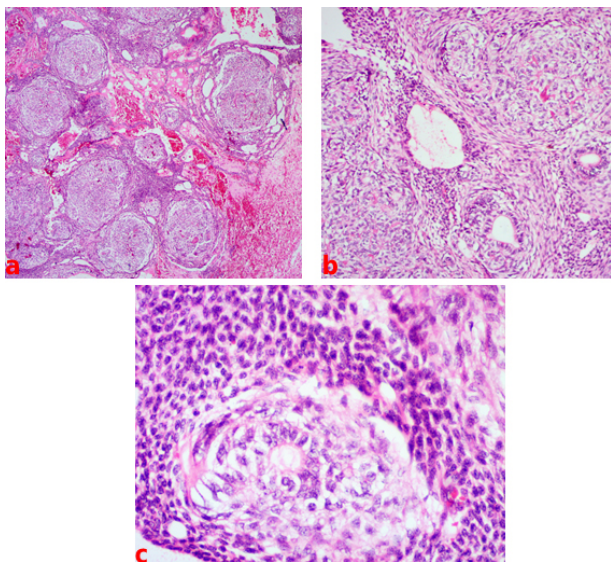


Figure 4. Histopathological assessment (H&E): **a)** a highly cellular tumor mass with the multinodular proliferation in the form of nests of rosette-like and duct-like structures formed by spindle, cuboidal, columnar, and Stellate reticulum-like cells (x40); **b)** a duct-like pattern of arrangement lined by cuboidal cells (x100); **c)** a single rosette with eosinophilic tumor droplets (x400).

A bone graft (Fig. 3c,d) was placed as the bony defect had limited regenerative potential [14] and persistence of this defect could affect the periodontal status of adjacent teeth #s 43 and 45. The enucleated mass was then analyzed radiographically (Fig. 5a,b). Patient follow-up was recommended to ensure bone healing and to detect signs of any recurrence or root resorption. The patient was examined one month after surgery; orthopantomogram which was taken to evaluate the treatment outcome revealed satisfactory healing (Fig. 5c).

The patient was scheduled for prosthetic rehabilitation in six months. The chronology of patient’s health events from day one to the postoperative period is depicted in Fig. 6.

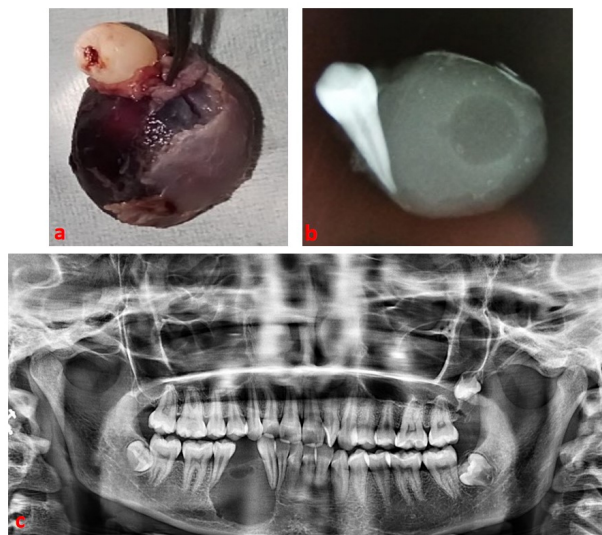


Figure 5. **a)** Enucleated specimen along with extracted tooth # 44; **b)** radiopaque flecks in the specimen on the radiograph; **c)** postoperative orthopantomogram showing missing tooth # 44 and surgical defect in the form of radiolucency one month after surgical treatment.

Discussion

Current WHO classifications categorize AOT as an epithelial odontogenic tumor, which accounts for 1% of cysts and tumors in the maxilla and 3% of other odontogenic tumors. AOT ranks fifth after ameloblastoma, cementoblastoma, odontoma, and myxoma [15]. There are three clinico-topographic variants of AOT [16–21], as illustrated in Table 1.

AOT is referred to as a “two-thirds tumor” as it occurs in the maxilla in about two-thirds of cases, about two-thirds of cases are diagnosed in young females, two-thirds of cases are associated with unerupted tooth, and two-thirds of affected teeth are canines [22]. Additionally, this tumor has been shown to occur on average between the ages of 3 and 82 years, with the distinctive feature that 68.6% of tumors are found in the second decade of life [23]. A literature review of 1, 558 reports of AOTs conducted by Chrcanovic *et al.* found that the average age of patients was 19.0 ± 9.0 years, indicating second-decade preponderance [11]. AOT is more common in the maxilla than in the mandible (2.1:1) [24]. A study conducted by Roza *et al.*, who reviewed 105 cases of AOTs and found that 55.1% of cases were seen in the maxilla [25], confirmed this. Similar findings were seen in a single-center study conducted in Sri Lanka, where 67.1% of cases were recorded in the maxilla and the remaining 32.8% of cases - in the mandible [4]. The tumors were predominantly seen in the anterior maxilla (48.5%), followed by the anterior mandible (29.5%), posterior mandible (15.2%), and posterior maxilla (6.6%) [25].

In 97% of intraosseous cases, follicular AOT is more common than extrafollicular AOT [26]. Extrasosseous or peripheral type is clinically restricted to gingival tissue and manifests itself as a mass projecting from the crown in 93% of cases [9]. According to the literature, the mandibular an-

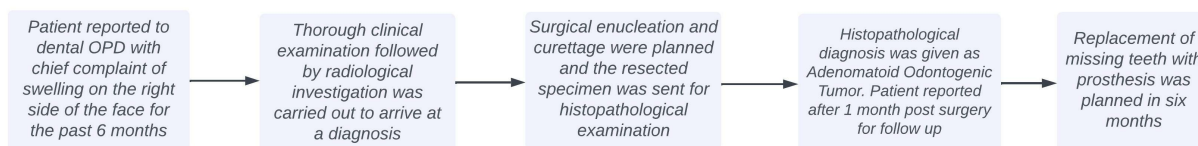


Figure 6. Timeline of events from day one to postoperative follow-up.

Table 1. Clinical variants of adenomatoid odontogenic tumor and their associated features [16–21].

Clinical variants of AOT	Associated Features	Prevalence
Follicular or pericoronal (intraosseous)	This type is associated with an unerupted tooth.	73%
Extrafollicular or extracoronal (intraosseous)	Extrafollicular tumors are further classified into four types: a) E1 - tumor with no relation to the tooth structure; b) E2 - tumor between two teeth roots producing apical divergence of adjacent roots; c) E3 - tumor superimposed on root apex; d) E4 - tumor superimposed on the mid-root level (intermediate part).	24%
Peripheral or extraosseous	Peripheral tumors are attached to the gingival structure with slight destruction of bone rarely distinguished on radiographs.	3%

terior labial gingiva is affected in 0.3% of cases only [15].

The extrafollicular variant mostly affects permanent canines [27] (89% of cases), with the occasional involvement of other permanent and deciduous teeth [28]. Becker *et al.* found that peripheral or extraosseous variants occurred only in 5% of cases. In addition, they noted that intraosseous tumors with extrafollicular variants were more commonly seen in the mandible than in the maxilla [26].

Bhagavathula *et al.* [29] described 16 AOTs, three of which were in the mandible. Each of the three cases involved an impacted tooth (#s 42, 43, 44, respectively), which manifested itself as swelling and obliterating buccal vestibule, accordingly. Walters *et al.* [5] reported a case of mandibular AOT with no impacted teeth in the anterior region and two more cases of mandibular AOT [30, 31]. Due to the absence of an impacted tooth associated with the tumor, which partially satisfies the two-thirds criterion, our case is identical to the case reported by Walters *et al.* [5]

Radiologically, AOT appears as a well-defined, unilocular, radiolucent lesion with a cyst-like sclerotic border. If the lesion affects the mandible, it seldom perforates the cortical bone, does not cross the midline, and has uneven boundaries [31]. In a review of published cases, Chrcanovic *et al.* discovered that bone expansion occurred in 89.5% of cases, whereas cortical bone perforation occurred in 45.6% of cases [11]. Moreover, they noted that the vast majority of cases (98.9%) were unilocular, with a clearly defined border in 98.7% of cases [11].

AOT, like ameloblastoma, can cause root resorption in rare situations. In a study by Fujita *et al.* [32], 8 cases of root resorption were reported. Premolar, molar, and adjacent anterior teeth commonly had root resorption, with an equal distribution in the maxilla and mandible. Except for one case in the mandible that was extrafollicular in type, all AOTs resulting in root resorption were of the follicular type. Bansal *et al.* documented one such case of aggressive AOT leading to root resorption of the adjacent anterior

tooth [33].

In 78.6% of cases, calcifications inside lesions appear as tiny, fine, uneven, distributed snowflakes or speckles [29]. These calcifications can be seen as calcified hypothesized regions, and radiopacities that may be fine and dispersed, discrete, irregular, or amorphous, being not visible in panoramic imaging [3]. The quantity and radiological method utilized influence the detection of these calcifications on the radiograph as well [3].

Similar to the radiographic findings mentioned above, this case likewise had radiopaque calcification with a distinct, well-defined corticated boundary. Ameloblastoma, dentigerous cyst, calcifying odontogenic tumor, central giant cell granuloma, calcifying epithelial odontogenic tumor, and keratocystic odontogenic tumor were among the potential diagnoses based on the radiographic findings [16]. Table 2 lists the distinguishing features of each of these conditions.

Multiple AOTs are a defining feature of Schimmelpenning syndrome, also known as sebaceous nevus syndrome. A linear nevus sebaceous, in addition to numerous neurologic, ocular, and skeletal abnormalities, is the most typical sign of this syndrome. These diseases seldom manifest intraorally, and when they do, benign odontogenic tumors and mucosal papillomatous development are the symptoms that are present. Two such cases of Schimmelpenning syndrome involving several AOTs have been reported in the literature [34, 35].

Conservative treatment is recommended as these tumors are benign, well encapsulated and have a low recurrence rate [36]. Curettage or enucleation of the tumor can be done while preserving the impacted tooth. Chrcanovic *et al.* observed a rather low recurrence rate (0.2%) with a mean follow-up period of around 30.03 ± 6.3 months [11].

Table 2. Differential diagnosis and its radiographic features [16].

Differential diagnosis	Radiographic features
Ameloblastoma	Unilocular or multilocular radiolucency frequently observed in the posterior mandible, either with or without an impacted tooth.
Dentigerous cyst	Follicular AOT continues to spread apically along the root past the cemento-enamel junction, whereas a cyst is related to the tooth at the cervical area.
Calcifying odontogenic cyst	A mixed lesion with radio-opaque flecks as its internal structure with well-defined boundaries. It occurs during the second and fourth decades.
Central giant cell granuloma	A multilocular lesion with thin wispy septa. In the majority of cases, cortication is not recognized.
Calcifying epithelial odontogenic tumor	A lesion is mostly located in the mandibular posterior region and affects people older than those commonly diagnosed with AOTs.
Keratocystic odontogenic tumor	A unilocular or multilocular radiolucent lesion with well-defined edges that are scalloped and corticated, septa having rough, curved edges, which occurs in the jaw posterior to the canine and may cross the midline.

Conclusions

The described clinical episode of an extensive tumor requiring surgical treatment highlights the various clinical, radiographic, and histopathological features of AOT. Although the extrafollicular variant is a rare entity, irrespective of its representation of two-thirds of tumors, gender prediction, location, and association with impacted and unerupted teeth, it should be included in the differential diagnosis of odontogenic tumors presenting as unilocular radiolucency as it is not always associated with unerupted or impacted teeth.

Patient Perspective

When I noticed a slowly growing swelling on the right side of my face for the past six months, I was concerned about it and reported it to a nearby dental hospital. I was examined thoroughly by doctors over there who took a detailed case history regarding the symptoms and performed necessary clinical and radiological investigations before arriving at a final diagnosis. I was assured that the swelling I had was a benign tumor that could be surgically treated with no further complications, given that I follow the postoperative instructions properly and show up for follow-up regularly. Surgical treatment with the removal of a tooth was done, after which I felt a little discomfort that resolved with the consumption of analgesics and antibiotics. I reported back after a month to check the response of the treatment received which revealed satisfactory healing.

Ethical Statement

This case report was conducted according to WHO Declaration of Helsinki – Ethical Principles for Medical Research Involving Human Subjects.

Informed Consent

Informed consent was obtained from the patient.

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Conflict of Interest

The authors declare that no conflicts exist.

Financial Disclosure

The authors did not receive support from any organization for the submitted work.

Data Availability

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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