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Adenomatoid odontogenic tumor: Clinical and radiological diagnostic challenges

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ABSTRACT

The adenomatoid odontogenic tumor (AOT) is a relatively uncommon odontogenic tumor with a relative frequency of 3–7%. The tumor is more common in females in their second decade of life. It exhibits a predilection for the maxillary anterior region. In this paper, we present a case of AOT in the mandible of a 27-year-old female. Histopathological examination of the tissue obtained from the initial incisional biopsy showed mostly an epithelial-lined cyst, which included a small nodule with the characteristic histological features of an AOT. Subsequent enucleation was done that confirmed the AOT diagnosis. The uncommon location of the lesion and the predominantly cystic configuration posed a clinical diagnostic challenge.

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1. Introduction

The adenomatoid odontogenic tumor (AOT) is a benign slow growing odontogenic lesion. The World Health Organization (WHO) stated that the lesion arises from the odontogenic apparatus odontogenic epithelium with mature fibrous stroma without ectomesenchyme.[1] It represents 3–7% of all odontogenic tumors.[1] Previously, the AOT was considered to be a variant of the ameloblastoma.[2] Histologically, due to the presence of duct-like structures, it is also considered as an adenoameloblastoma.[3]

The literature review was done on documented cases of adenomatoid odontogenic tumor in the mandible. PubMed database was used to search published research reports using the terms ''Adenomatoid Odontogenic Tumor'' AND ''Mandible''. A total of 58 publications were identified from the search report (see Fig. 1). However, 23 publications were suitable for data collection based on inclusion and exclusion criteria. The inclusion criteria for selecting research papers were the following: full-text paper availability, written in English language, adequate and clear content on location, and diagnostic details of adenomatoid odontogenic tumor. The exclusion criteria were the following: incorrect web-link, title and/or abstract duplication, and general review papers. The data were analyzed for specific location in mandible (anterior or posterior), type of tumor (extrafollicular, follicular, or peripheral), information on unerupted/impacted tooth, clinical and radiological diagnosis, and concurrent odontogenic pathologies reported. Thirty-five papers were excluded due to no access to full-text of the research papers, and further two papers were excluded as the content did not match our search criteria (Table 1).

Twenty-one published reports constituted a total of 145 cases of mandibular adenomatoid odontogenic tumor; there were 89 cases of anterior mandible[4–16], 48 cases of posterior mandible[4,17–23], and 8 cases of mandibular adenomatoid odontogenic tumor that did not specify the exact location.[24]

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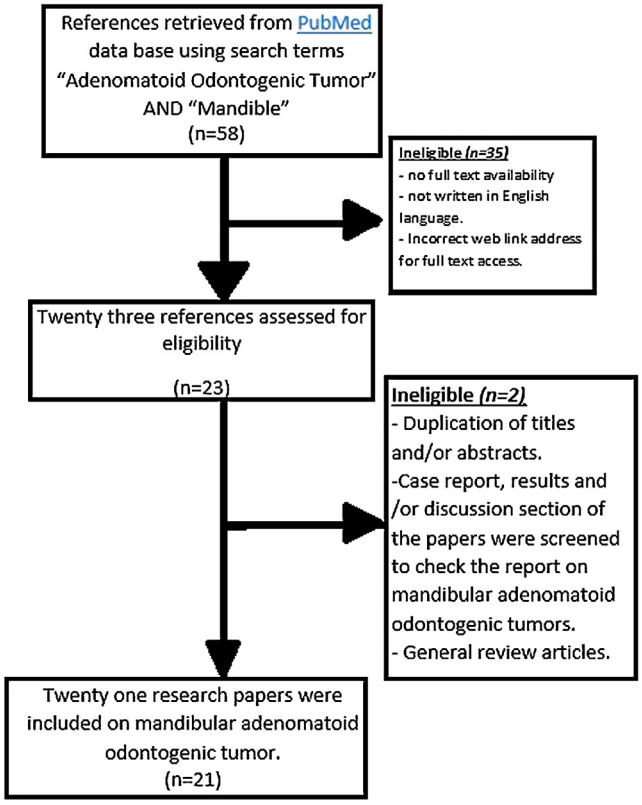


Fig. 1 – Flow chart showing the selection procedure of research papers.

Seventy-four cases were follicular type,[4,5,8–10,15–19,22,23,25] 49 cases were extrafollicular[4,6,7,14,17,18,20,21], 3 cases were peripheral,[11,18] 11 cases were reported as intraosseous[13], and 8 cases did not provide details on type.[24] Four published reports mentioned an unerupted or impacted tooth (lateral incisor, canine, or premolar).[5,6,22,23] Fifteen published reports mentioned clinical and/or radiological diagnosis. The most common clinical and/or radiological diagnosis made was dentigerous cyst (5 cases).[5,9,15,17,23] Other diagnoses included adenomatoid odontogenic tumor (4 cases),[10,13,18,19] ameloblastoma (2 cases),[8,14] residual cyst (1 case),[21] odontogenic keratocyst (1 case),[7] gingival epulis (1 case),[11] and odontogenic cyst (1 case).[16] Three published papers reported concurrent odontogenic pathologies and that included dentigerous cyst (1 case),[22] odontoma (1 case),[20] and cemento-ossifying fibroma (1 case).[9]

In this paper, we present a case of AOT in an uncommon location and which histologically showed the presence of a significant amount of dentigerous cystic epithelium.

2. Case report

A 27-year-old female Jamaican patient of African descent visited a dental office with a chief complaint of swelling in the anterior mandibular region (Figs. 2 and 3) for the past 9 months. The swelling was asymptomatic and her oral hygiene was good. The intraoral examination revealed a nontender

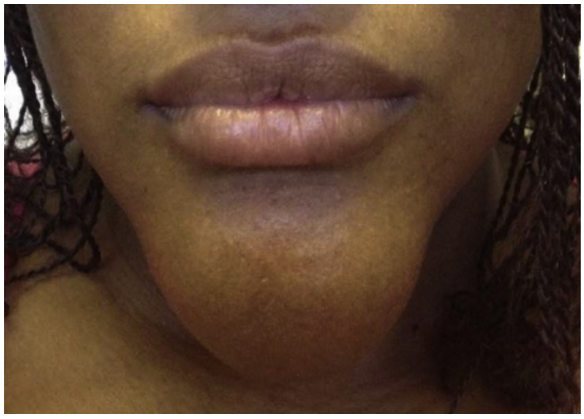


Fig. 2 – Swelling in the mandibular anterior region.

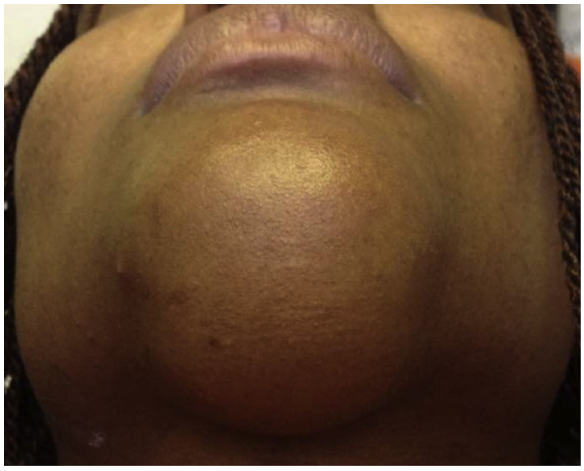


Fig. 3 – Extent of swelling in anterior mandibular region (submental view).

labial and lingual expansion of the mandibular anterior cortical plates, which were covered by normal appearing mucosa. The patient had no nerve deficit or adenopathy in the facial or cervical region. An orthopantomograph revealed the presence of a significantly large unilocular radiolucency with ill-defined borders involving the mandibular anterior region and encircling the unerupted left mandibular canine that had radiopaque areas of varying sizes and shapes that resemble calcified deposits. The unilocular radiolucent area was observed to extend from the left first premolar to the right first premolar area. Displacement of the left and right central, lateral incisors as well as retained left deciduous canine was seen. Root resorption of the mesial aspect of the apical root of the left first premolar was also noted (Fig. 4). Based on the clinical and radiological findings, the lesion was diagnosed as a dentigerous cyst with a differential diagnosis of ameloblastoma.

An initial incisional biopsy of the lesion was performed. Microscopically, the hematoxylin- and eosin-stained tissue of the initial biopsy demonstrated mostly a cyst, with a well-defined fibrous capsule and a lining mostly of thin non-keratinized stratified epithelium. A small lesion comprised of

Table 1 – Comparison of Clinical, radiological characterstics and concurrent pathologies from published reports.

Description	Type of Adenomatoid odontogenic tumor	Clinical/ radiological diagnosis	Concurrent association of other pathologies	Study
Anterior mandible [20 cases] Posterior mandible [7 cases]	Follicular type: 14 Extrafollicular type: 13	–	–	Adisa et al. (2016)[4]
Premolar region: 1 case	Follicular type (unerupted premolar)	Dentigerous cyst	–	Belgaumi et al. (2015)[23]
Anterior mandible: 1 case	Follicular type (unerupted lateral incisor)	Dentigerous cyst	–	Seo et al. (2015)[5]
Posterior mandible [1 case] in 19-year-old female	Follicular type (unerupted first premolar)		Dentigerous cyst	Manjunatha et al. (2012)[22]
Posterior mandible [1 case] in 18-year-old male	Extrafollicular type	Residual cyst	–	Shivali et al. (2013)[21]
Anterior mandible [4 cases] in a 20-year-old female, 13-year-old female, 13-year-old female, and 24-year-old female	2 extrafollicular type and 2 follicular type (unerupted canine)	–	–	Mohanty et al. (2013)[6]
Anterior mandible [1 case] in 17-year-old male	Extrafollicular type	Odontogenic keratocyst	–	Saluja et al. (2013)[7]
Posterior mandible [1 case] in 32-year-old male	Extrafollicular type	–	Odontoma	Gomez et al. (2013)[20]
Posterior mandible [1 case] in 14-year-old male	Follicular type	Adenomatoid odontogenic tumor	–	Bhatt et al. (2013)[19]
Anterior mandible [1 case] in 17-year-old female	Follicular type	Ameloblastoma	–	Narayanan et al. (2013)[8]
Anterior mandible [1 case] in 30-year-old female	Follicular type	Dentigerous cyst	Cemento-ossifying fibroma	Prakash et al. (2012)[9]
Anterior mandible [46 cases] Posterior mandible [33 cases]	Pericoronal type: 48 cases Extracoronal type: 30 cases Extraosseous type: 2 cases	Adenomatoid odontogenic tumor	–	Becker et al. (2012)[18]
Anterior mandible [1 case] in 14-year-old male	Follicular type	Adenomatoid odontogenic tumor	–	Sharma et al. (2012)[10]
Anterior mandible [1 case] in 10-year-old male	Extraosseous type	Gingival epulis	–	Kumar et al. (2012)[11]
Posterior mandible [1 case] in 24-year-old female	Follicular type	Dentigerous cyst	–	Bhullar et al. (2011)[17]
Anterior mandible [1 case]	–	–	–	Varkhede et al. (2011)[12]
Anterior mandible [9 cases] Posterior mandible [2 cases]	Intraosseous type [11 cases]	Adenomatoid odontogenic tumor	–	Saghravanian et al. (2010)[13]
Anterior mandible [1 case] in 15-year-old female	Extrafollicular type	Ameloblastoma	–	Yilmaz et al. (2009)[14]
Anterior mandible [1 case] in 9-year-old female	Follicular type	Dentigerous cyst	–	Vera Sempere et al. (2006)[15]
Anterior mandible [1 case] in 23-year-old male	Follicular type	Odontogenic cyst	–	Handschel et al. (2005)[16]
Mandible [8 cases]	–	–	–	Ajayi et al. (2004)[24]

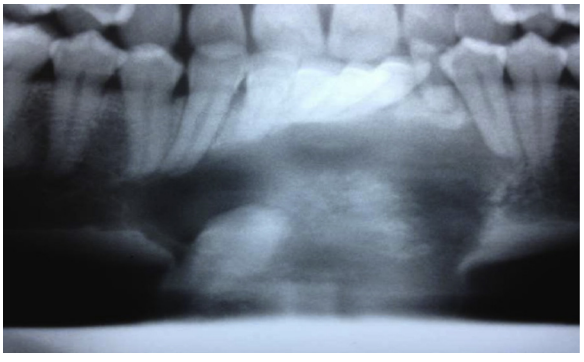


Fig. 4 – Unilocular pericoronal radiolucency with ill-defined borders. Note the presence of radiopaque mass, displacement of mandibular anterior teeth, and resorption of left mandibular canine.

nodular aggregates of epithelial cells arranged in different patterns, including duct-like structures, was seen within the wall. Based on these findings, a diagnosis of AOT was made. The diagnosis of AOT was confirmed as the enucleated specimen contained an abundance of epithelial tissue in the form of nodular aggregates of epithelial cells, some of which were spindle and ducts, and some contained eosinophilic material (Figs. 5 and 6). Dystrophic calcification was abundant in the lesional tissue and arborizing trabeculae of the odontogenic epithelium was prominent at the periphery. However, in the original biopsy, a significant amount of the cyst had thin nonkeratinizing stratified squamous epithelium, as seen in dentigerous cysts.

Following the histopathologic report, enucleation of the lesion was done under general anesthesia. The mandibular

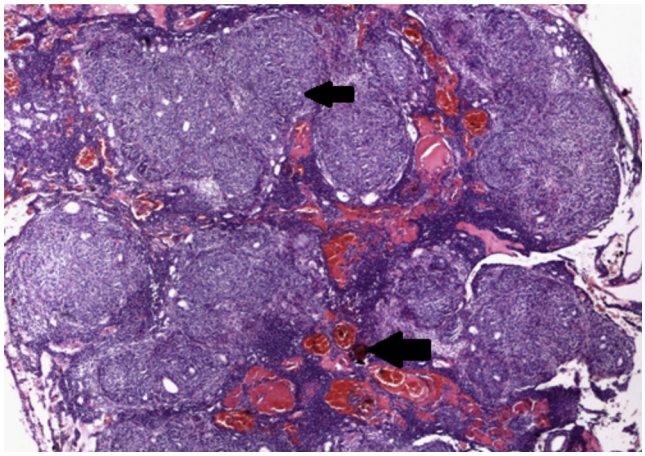


Fig. 5 – Low-power photomicrograph showing nodular aggregates of cells. Note a small annular calcific deposit in the lower half of the picture (hematoxylin and eosin stain).

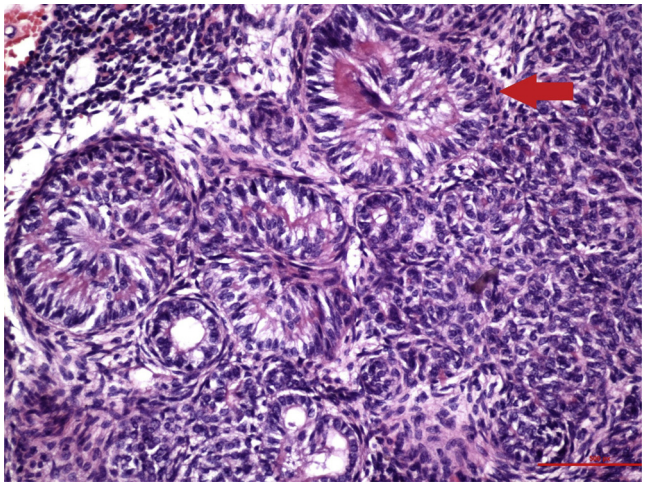


Fig. 6 – High-power photomicrograph showing gland-like structures with focal eosinophilic hyaline material in one such structure in the upper half of the picture (hematoxylin and eosin stain).

right second premolar, first premolar, and canine, along with the mandibular left and right central and lateral incisors, impacted mandibular left canine, and retained deciduous teeth were removed due to the compromised periodontal status and impaction. Prosthetic replacement (removable/fixed) was included in the treatment planning. The one-year postoperative visit showed no signs of recurrence.

3. Discussion

The AOT is a slow growing, benign odontogenic tumor usually occurring in the anterior maxilla and has a female predilection.[26,27] In our case, the lesion occurred in the anterior mandible, which is a less common location. Clinical features generally center on patient complaints regarding a missing tooth and tooth displacement. Unless it is secondarily infected, the lesion usually presents as an asymptomatic swelling, which is slow growing and is often associated with an unerupted tooth. Cortical plate expansion, however, sometimes causes pain. These contribute to the diagnosis.[3] The AOT occurs in both intraosseous and peripheral forms. Radiographically, the intrabony variants comprise follicular and extrafollicular types. The follicular type is associated with an unerupted tooth and mimics a dentigerous/follicular cyst.[26] In contrast, the extrafollicular type is not associated with an unerupted tooth. It can be found between roots, or above, or superimposed on erupted permanent teeth roots. The present case had an unerupted permanent left mandibular canine and presented as an asymptomatic swelling.

The AOT is sometimes referred to as the two-thirds tumor, as it occurs in the maxilla in about 2/3 of cases, about 2/3 of AOT cases are observed in young females, 2/3 cases are associated with an unerupted tooth, and 2/3 of the affected teeth are canines.[1] Dentigerous cyst, unicystic ameloblastoma, calcifying epithelial odontogenic tumor, calcifying odontogenic cyst, and ameloblastomic fibro-odontoma are considered in the panel of radiological differential diagnosis of adenomatoid odontogenic tumor. A radiographic differential diagnosis of dentigerous cyst is considered in AOT cases where there is a radiolucency enveloping the crown of an unerupted tooth. However, the differential diagnosis of dentigerous cyst is excluded in AOT cases where the radiolucency is extending beyond the crown–root junction.[18,23,28] While the unilocular radiographic appearance may simulate the appearance of unicystic ameloblastoma,[8] opaque areas (calcifications) in the radiographic interpretation may give an appearance of calcifying epithelial odontogenic tumor,[29] calcifying odontogenic cyst,[30–33] or ameloblastic fibro-odontoma.[30] However, sometimes adenomatoid odontogenic tumor may present as multilocular radiolucent areas. In such instances ameloblastoma or odontogenic keratocyst may be considered as the radiological diagnosis.[34]

A diagnosis of AOT should be considered in the radiological differential diagnosis of pericoronal radiolucencies, especially in teenagers and young adults with an impacted tooth, if radiopaque foci are identified. If there are no flecks of radiopacities, then a dentigerous cyst is more likely.[10] However, dentigerous cyst and ameloblastoma do not usually reveal calcification in radiographs although they can present as unilocular pericoronal radiolucency associated with impacted teeth. A radiological differential diagnosis of pericoronal radiolucencies with radiopaque foci should consider the following lesions: calcifying epithelial odontogenic tumor, ossifying fibroma, cemento-ossifying fibroma, and ameloblastic fibro-odontoma. Further, these radiopaque foci of calcification are more clearly visualized in an intraoral periapical radiograph than in an orthopantomograph.[1] The present case showed a pericoronal radiolucency surrounding the unerupted left mandibular canine. However, the central area of the lesion showed a radiopaque mass that resembled calcification.

An interesting histopathological finding seen in the present case was the presence of a significant amount of nonkeratinized stratified squamous epithelium, as seen in a dentigerous cystic lining. Similar cases have been recently published, with one of an Asian individual being similar to this one.[7,35]

The nature of the AOT is controversial. Some authors support the theory that AOT is a hamartomatous growth while others consider it to be a neoplastic growth with sound arguments advanced in favor of and against both hypotheses.[35] The dental lamina remnants are likely to represent the progenitor cells. The World Health Organization in 2005 defined AOT as a tumor composed of odontogenic epithelium presenting a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth.[10]

Interestingly, the present case had some unusual clinical, radiographical, and histological features that distinguished it from conventional AOT. Firstly, AOT occurs predominantly in the anterior maxillae and is relatively small in size, usually up to 3 cm in size.[36] In contrast, the present case was seen in the anterior mandibular region and was greater than 3 cm in size. The size estimated clinically was between 3.5 and 4.0 cms. Similar mandibular involvement was observed in other patients.[9,10,37,38] Secondly, radiographic examination usually reveals a well-circumscribed radiolucency with an impacted tooth, but the present case showed a large radiolucent defect with ill-defined borders, radiopaque areas, and root resorption. To our knowledge, only 5 cases of AOT with root resorption have been noted.[39–43] Thirdly, much of the tissue examined showed a simple epithelium-lined cyst, but where present, the histologic features of AOT were distinctive.

The present case was treated by surgical enucleation. Most cases of AOT are benign in nature, with conservative enucleation or curettage being the treatment of choice. A report from the Japanese literature suggests a recurrence rate of 0.4%.[42]

4. Conclusion

A diagnosis of AOT should be considered in the radiological differential diagnosis of pericoronal radiolucencies, especially in teenagers and young adults with an impacted tooth, if radiopaque foci are identified. If there are no flecks of radiopacities, then a dentigerous cyst is more likely. Radiopaque foci of calcification are more clearly visualized in an intraoral periapical radiograph than in an orthopantomograph. The present case report supports the importance of histopathological examinations of all surgically removed tissues.

Conflicts of interest

The authors have none to declare.

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