

Mandibular Adenomatoid Odontogenic Tumor: A Rare Entity

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ABSTRACT

An adenomatoid odontogenic tumor (AOT) is a very rare epithelial tumor that makes up to 3% of odontogenic tumors. The mandibular region accounts for 35% of cases, while the maxilla accounts for around 65% of cases. This case report aims to report a case of adenomatoid odontogenic tumor in the anterior mandible in a male patient who was treated with surgical marsupialization and posterior enucleation. Auxiliary therapeutic techniques can be used effectively to reduce the lesion, and accurate histological diagnosis is mandatory to avoid unnecessary aggressive surgery. This case study also covered the need for a thorough description that distinguishes AOT from other odontogenic tumors, of which dentigerous cyst is the most frequent misdiagnosis.

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Introduction

Adenomatoid odontogenic tumor (AOT) is a rare odontogenic tumor of epithelial origin that arises mostly from the enamel, enamel epithelium, dental lamina, or the Malassez rests. This is currently the most accepted theory.¹ Most of the time, it is misdiagnosed as an odontogenic cyst or a dentigerous cyst. Earlier known along a wide range of nomenclature, such as adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinoma, or teratoma odontoma. It was in 1905 that Steensland described AOT for the first time.² Later in 1999, Philipsen et al presented reports and a review based on literature gathered up to 1997. This showed interesting and distinguished features of AOT.³

Histopathologically, WHO described AOT as an

epithelioid tumor with duct-like structures and connective tissue stroma. Tumor shows a partial cystic component in a few cases; mostly, solid lesions are noted. Eosinophilic, uncalcified, amorphous material called tumor droplets is present. This may be in the form of plaques or homogeneous material, highly useful during diagnosis.⁴

Immunohistologically, the AOT phenotype is characterized by cytokeratin (CK): CK5, CK17, CK19. Criveline detected CK 14, which shows origin from reduced dental epithelium. Tekatrashi et al observed positive staining for iron-binding proteins (transferrin/ferritin) and proteinase inhibitors (alpha one anti-trypsin).⁵

Radiographically, intra-oral periapical radiograph seems to be more useful than panoramic view as discrete foci with flocculants pattern with minimal

small calcifications are visible. The classical picture is of a well-circumscribed, radiolucent lesion, causing no cortical bone erosion or thinning, having a sclerosing border. Most cases have calcifications within them, which may not be visible in radiographs. It may present as a single lesion, a large cystic lesion, or multiple cystic masses. This leads to common misdiagnosis of dentigerous cyst, calcifying odontogenic cyst, calcifying epithelial odontogenic tumor, ameloblastoma, and odontogenic keratocyst. MRI scans help to distinguish AOT from other odontogenic tumors.⁶

Clinically, the male-to-female ratio is 1:2, predominantly in the Asian population. Delayed eruption of a permanent tooth, slowly disfiguring facial appearance at the lower jaw, and painless swelling are the common presentations.⁷ This case report shows the distinct characteristic features of AOT in a 16-year-old boy.

Case Presentation

A 16-year-old patient was referred from the oral medicine department with chief complaints of a swelling, progressively increasing in size at the lower left jaw, near the para-symphysis mandible, for the last 5 months. The patient also complained of a mobile deciduous tooth on the left side. An orthopantomogram was requested, which showed a unilocular, radiolucent swelling with a sclerotic border at the para-symphysis of the mandible, attached with an unerupted tooth. The OPG showed unilocular cystic swelling with an unerupted tooth and a deciduous tooth at the carina. With clinical and radiological findings, an initial diagnosis of dentigerous cyst was made. Decision to carry out Marsupialization followed by enucleation of the cyst was made and therefore operated through an intra-oral approach. The cyst lining, along with the deciduous teeth at the carina, was extracted and sent for histopathological analysis. The cystic cavity was washed with normal saline and then packed with the polyfax-soaked gauze. In a follow-up visit

after 1 day, the cavity was washed, and the dressing was changed. After one week patient was referred to the prosthodontic department for the impression of a prosthetic plug. Within two days prosthetic plug was ready and placed.



Figure 1: Pre-operative clinical picture of a swelling, noted near the left mandibular para-symphysis



Figure 2: Intra-Oral picture



Figure 3: Pre-operative OPG



Figure 4: Aspiration Fluid



Figure 5: Per Operative with dressing



Figure 6: Intraoperative Illustrations



Figure 7: Post Operative Radiograph

Histopathology report revealed an Adenomatoid Odontogenic Tumor (AOT) measuring 1.5×0.5×0.5 cm and filled with keratinous debris. After this Histopathology report, a treatment plan for excision of the tumor was made and discussed with the patient. The lesion was surgically enucleated under general anesthesia. Macroscopically, the mass was well encapsulated with cystic areas along with an embedded permanent mandibular left canine in the tumor mass. Histopathological examination revealed sheets, ducts, and whorls of darkly staining ovoid to round epithelial cells, suggestive of odontogenic epithelial cells. The duct-like structures were lined by columnar cells. A few basophilic calcifications were also observed. Small cystic areas

containing degenerated cell debris were noted in the focal areas. The supporting connective tissue stroma was loose and less cellular in nature. Based on these findings, a histopathologic diagnosis of adenomatoid odontogenic tumor was made. The patient was under follow-up and had not shown any signs of recurrence six months after surgery.

Discussion

The histopathological features of AOT include odontogenic epithelium in the shape of sheets or rods and a small number of odontogenic cells arranged in a duct-like pattern with eosinophilic substance inside. At the edges, there is a distinct fibrous tissue capsule.⁸

Our histopathological report showed spindle-shaped stroma with glandular structures lined by columnar epithelium with an unerupted tooth, suggestive of a follicular type of AOT, which accounts for 73% of all cases. AOT is most commonly misdiagnosed as a dentigerous cyst due to its common areas of development and similar radiographic picture. In our case, the anterior portion of the mandible was affected, in association with an unerupted tooth and a displaced permanent tooth. Radiographically, in a panoramic view, it appears as a circumferential, volumetric, radiolucent, and unilocular lesion with multiple calcifications.⁹ Symptomatically, it is a non-tender, progressive lesion causing cosmetic deformity with no involvement of adjacent structures. AOT is classified under hamartoma, meaning it has limited potential for growth and is a developmental anomaly of tissue differentiation. AOT is called a two-thirds tumor as two-thirds of cases occur in females, two-thirds of cases with an unerupted tooth, two-thirds of cases occur in the maxilla, and in two-thirds of cases, the Canine is the affected tooth.¹⁰

Our 16-year-old patient had the characteristic features. Swelling was noted at the parasymphysis of the mandible with an unerupted

tooth. The tooth involved was the left canine. The cyst measured around 2.8×1.8×1.5 cm. Generally, AOT measures around 1-3 cm. Therefore, it fits into the criteria. Since AOT does not infiltrate the adjacent bones and treatment mainly is enucleation, therefore most cases, it does not recur. Radiopacities and calcifications are present within the lesion, which is particularly seen only in AOT.¹¹ A dentigerous cyst does not have calcifications within it. Also, radiographically, AOT is shown to involve coronal and radicular aspects of the tooth, but a dentigerous cyst only surrounds the coronal aspect of the involved tooth.¹² AOT does not cause resorption of the root of the tooth involved. In most cases, it causes displacement of the teeth. Canines are most commonly involved, followed by premolars and rarely molars.¹³ The variants extra follicular type has an intra-bony lesion and no connection with the tooth. Peripheral variant presents as a gingival swelling, small sessile masses on the buccal gingiva. Both variants are only diagnosed histologically as they involve gingival pathologies.¹⁴

Conclusion

The differential diagnosis of AOT with other pathologies, such as ameloblastoma, keratocyst, or a dentigerous cyst, is important due to the aggressive treatment they require. AOT requires surgical conservative treatment as they do not involve the bone. The case report provides detailed descriptive features of AOT for accurate diagnosis and treatment. Delays in diagnosis leads to an increase in the size of the lesion and, therefore, a larger area of clearance is needed.

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