

Adenomatoid Odontogenic Tumor of mandible – A Case Report

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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.

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) was first described by Steensland in 1905 [1]. Unal et al [2] produced a list of nomenclature for AOT reported in literature, they are adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantium or teratomatous odontoma and is currently called as AOT. Dreibaldtin in 1907 described it as a pseudoameloblastoma [3]. Harbitz in 1915 [4], reported it as cystic adamantoma. Ghosh in 1934 described it as adamantinoma and was recognized by Stapne in 1948 as a distinct pathological entity. Finally in 1969 Philipsen and Birn⁴ proposed the term AOT and was adopted by WHO in 1971 [5].

According to the second edition of the World Health Organization journal 'Histological Typing of Odontogenic Tumors', AOT is defined as 'A tumor of odontogenic epithelium with duct-like structures and varying degrees of inductive change in the connective tissue. The tumor may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst [6].

It is an uncommon odontogenic tumor with frequency to 7.1% [7]. It appears as an intraoral and extraoral swelling. It is referred as 'Two-third tumor' because it occurs in maxilla in about 2/3 cases, about 2/3 in young females, 2/3 cases are associated with unerupted tooth and 2/3 affected teeth are canines [5]. This report describes an AOT in the mandible, illustrate the clinical, radiological and microscopic features.

CASE REPORT

A 22-year-old female patient came with the chief complaint of a swelling in the lower front teeth region since 8 months. She noticed a swelling in the lower chin region 8 months back and it's the same since then. She has no relevant medical or dental history except for pain on pressure over the swelling. On examination, there was a single swelling in the chin region, measuring about 7 × 5 cm, ovoid in shape with smooth surface, extending from the midline 3 to 4 cm posteriorly and 4 mm from the corner of the mouth till 1 cm beyond the inferior border of the mandible supero- inferiorly with mild facial asymmetry due to the swelling in the left side chin region. On palpation, it was bony hard, well defined and tender.

Intraorally, 33 was missing and there was a swelling involving the buccal and lingual cortical plates extending from 34 to 42 region buccally and 34 to 42 region lingually. On palpation, it was bony hard except in relation to 33 region where it was yielding, well defined and tender on palpation. All the teeth in the region of the lesion were vital. Aspiration was negative.

Considering the clinical features, the lesion was provisionally diagnosed as a dentigerous cyst and probable differential diagnosis can be any benign odontogenic tumor.

Intraoral periapical radiograph revealed horizontally impacted 33, surrounded by a well-defined radiolucency and floccular radiopacities around the crown of the impacted 33. Lingual bone is thinned. Orthopantomogram revealed a single unilocular, well defined radiolucency on the midline of the mandible with hyperostotic border, measuring 7 × 5 cm, oval in shape, extending mediolaterally from apical region of 42 to 34 and superoinferiorly 0.5 cm from crest of the alveolar bone till above the inferior border of the mandible. The radiolucency completely encloses the tooth including root. The internal structure revealed presence of radiopaque structure morphologically similar to that of a tooth extending from center of radiolucency obliquely toward the border, presence of a floccular radiopacity around the crown of the tooth structure. There is external resorption in relation to 41, 31, 32 with distal displacement of the roots. With the evidences of the radiographs, the provisional diagnosis can be modified to an adenomatoid odontogenic tumor or a calcifying epithelial odontogenic cyst. Hematological, biochemical and urine analysis were done to rule out any systemic causes and all were normal.

Histopathological examination revealed polygonal cells with focal areas of eosinophilic coagulum, hematoxyphilic globular calcifications and hematoxyphilic irregular calcifications. The polygonal cells show tightly packed whorled arrangement. The fibrous connective tissue exhibits cords, islands and sheets of polygonal cells.

After all the required investigations were done an intentional root canal therapy of 34 was planned followed by curettage and enucleation of the lesion. Histopathological examination of the enucleated lesion confirmed the diagnosis of AOT.



OPG showing circumscribed radio-lucent lesion with calcifications and impacted teeth



DISCUSSION

AOT usually occurs within the tooth bearing areas of jaws and often found in association with impacted teeth. The origin of AOT is controversial, but many authors believe in odontogenic source. AOT has cytological features similar to various components of enamel organ, dental lamina, reduced enamel epithelium, and its remnants.[8] Radiographically, AOT frequently looks like a dentigerous cyst. The lesion is usually unilocular and radiolucent. However, they contain fine calcifications (snowflake), a feature that may be helpful in differentiating an AOT from dentigerous cyst. The unilocular radiolucency is well demarcated with smooth cortical border. Most lesions are pericoronal, juxta coronal, and divergence of roots and displacement of teeth often occurs without root resorption.[9,10,11,12]

SUMMARY AND CONCLUSION

Even though enucleation and curettage for AOT is the most common treatment modality, accurate histological diagnosis is mandatory to avoid unnecessary mutilating surgery. Still the search for accurate classification and ideal nomenclature for AOT continues. The debate as to whether AOT is an anomalous hamartomatous growth or a true benign neoplasm has not been settled yet.[13] Immunohistochemical studies by certain authors reinforce the theory of hamartomatous character of this lesion indicating AOT is not a true neoplastic lesion.[14] Some authors prefer to disagree with the term AOT.[15] According to Marx and Stern, the more appropriate term is adenomatoid odontogenic cyst (AOC).[15] The term adenomatoid odontogenic cyst as suggested by Marx and Stern is controversial. But in our case presented, the presence of unilocular cystic lesion, fluid on aspiration, and cystic cavity on transection has to some extent support the terminology adenomatoid odontogenic cyst (AOC) as termed by Marx and Stern.[15]

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