Adenomatoid Odontogenic Tumor: An Unusual Presentation

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Abstract

Adenomatoid odontogenic tumor (AOT) is a not so common neoplasm of the odontogenic origin, accounting for around 3-7% of all odontogenic tumors. It is usually present in young females in the anterior maxillary region and thus commonly referred to as a two-third tumor. It is a benign (hamartomatous), usually non-invasive tumor which shows slow and progressive growth. Due to its rarity and varied clinical presentation it is often misdiagnosed. To add to confusion further, there are various lesions which mimic AOT closely. Herein this article, we present a case report of a young female patient with an unusual presentation of AOT tumor in the mandibular region focusing on the versatility of this rare entity.

Keywords: Adenomatoid odontogenic cyst, Adenomatoid odontogenic tumor, Hamartoma, Odontogenic neoplasm, Two-third tumor

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a benign, non-aggressive, gradually progressive lesion which has been known to dental fraternity since more than 100 years. Credit for the first description of the lesion goes to Steensland.¹ Since then, many clinicians have reported it under different names. Unal et al. compiled the list of all the synonyms of AOT, which included adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum, or teratomatous odontoma.² In 1969, Philipsen and Birn coined the current nomenclature, i.e. AOT.³ In 2005, the second edition of World Health Organization histological typing defined it as, “a tumor of odontogenic epithelium with duct like structures and with varying degrees of inductive changes 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.”⁴ Due to its usual clinical presentation, it is commonly referred to as “two-third tumor.” The two-third cases are seen in young females in the second decade of life. The two-third cases are seen in maxilla and most often associated with an impacted canine.⁵-⁷ However, the unusual presentation as seen in our case validates it to be called as “Ruler of disguise.” The striking features which make our case report unique are as tabulated in Table 1.

CASE REPORT

A 14-year-old female patient presented with swelling of lower 3rd of the face on the right side since 3 months. Initially, the swelling was small and has gradually increased in size to the present status. No history of associated pain or discharge from the swelling was present. Besides this, the patient also gave a history of decayed right lower back primary tooth since 6 months that was mobile from last 10-15 days and was exfoliated 2 days back. Nothing relevant in medical history, personal, or family history was evident. On extra-oral examination,
a single, diffuse swelling extending from the corner of mouth to the inferior border of the mandible, measuring approx. 4 cm × 3 cm in dimensions was seen. The overlying skin was slightly stretched with no discharge and discoloration. On palpation, it was non-tender, firm with well-defined borders (Figure 1a). Intraoral examination revealed a single, oblong shaped swelling extending from 43 to 45 in right buccal vestibule causing obliteration of vestibule and bi-cortical expansion. Mandibular right first premolar was missing, and mobility was present in right mandibular canine (Grade I), and right mandibular second premolar and molar (Grade II). Drifting of adjacent teeth was noticed (Figure 1b).

On the basis of the acute history of swelling associated with the decayed tooth, a working diagnosis of the infected radicular cyst was made, and the patient was subjected to further investigations. Radiographic examination consisted of an intraoral periapical radiograph of right mandibular premolar region, mandibular occlusal radiograph and orthopantomograph. It showed a well-defined, unicystic, unilocular radiolucent lesion in relation to impacted right mandibular first premolar. It measured approximately 3.5 cm × 2.2 cm in widest dimensions. The borders of the radiolucency were well-defined, smooth, and corticated. The mandibular right first premolar was impacted and was located at the floor of the lesion. The internal structure revealed multiple flecks of radiopacities along with an impacted tooth. There was thinning and cortical perforation of the inferior border of the mandible. The lesion resulted into the divergence of mandibular right first premolar and molar (Figure 2a-c). Radiographic diagnosis was suggestive of AOT with differential diagnosis of calcifying epithelial odontogenic tumor (CEOT) and dentigerous cyst. Aspiration using wide bore needle was done which consisted a copious amount of clear rust colored fluid, approximately 2 ml without any crystals (Figure 3a).

The incisal biopsy was done, and histopathologic examination revealed cuboidal or columnar epithelial cells forming nests or rosette like structure with the central eosinophilic amorphous material. Varying sized duct like spaces lined by low columnar cells were present within the nodules. Fragments of crystalline calcification resembling cementum and amyloid like material were also seen (Figure 3b).

Thus, a final diagnosis of AOT was confirmed.

**DISCUSSION**

AOT is slow-growing lesion accounting for 0.1% of all odontogenic tumors. It has an affinity for anterior maxilla.
associated with impacted canine. The young female patients in 2nd-3rd age group constitute the most common patient population. In our case, it was seen as swelling in the lower jaw which was unusual. In the majority of cases, it manifests as non-aggressive lesion which accounts for painless swelling. However, in our case, it was associated with bicortical expansion and perforation of the mandibular cortex.

Philipsen et al categorized AOT as follicular, extrafollicular, and peripheral. The follicular type (74%) is a central intraosseous lesion associated with an impacted tooth while extrafollicular intraosseous AOT (24%) has no relation with an unerupted tooth. The peripheral variant (3%) appears as a gingival fibroma or epulis attached to the gingiva.

Radiographically it presents as a unilocular radiolucent lesion associated with impacted tooth and radiopaque foci. However, multicocular large radiolucent lesions have also been reported in the literature. Tooth displacement is a common finding than root resorption. It should be differentiated from similar appearing conditions such as dentigerous cyst, CEOT, calcifying odontogenic cyst, ameloblastoma, keratocystic odontogenic tumor, globulomaxillary cyst, ameloblastic fibro-odontoma, and intermediate-stage odontoma. Dentigerous cyst appears as pericoronal radiolucency attached to cementoenamel junction. While AOT envelopes both the crown and root portion. CEOT is more common in posterior mandible in older age group. In CEOT, varying sized radiopaque foci are clustered in vicinity to the crown, thereby giving typical ‘driven snow’ appearance.

Treatment for AOT is surgical excision. Recurrence rate is low (0.2%) because of its non-aggressive behavior, and presence of capsulated lesion.

**CONCLUSION**

We conclude that the though uncommon but the two-third tumor should be kept in mind while listing the possible causes of swelling occurring in anterior mandibular region.

**REFERENCES**


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