CASE REPORT

Adenomatoid odontogenic tumour: An enigma

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KEYWORDS

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Abstract

Adenomatoid odontogenic tumour (AOT) is an uncommon, hamartomatous, benign epithelial lesion of odontogenic origin. The current World Health Organisation (WHO) classification of odontogenic tumors defines AOT as being composed of the odontogenic epithelium in a variety of histarchitectural patterns, embedded in mature connective tissue stroma, and characterized by slow, but progressive growth. The aim of this paper is to present three rare cases of intraossous AOTs with varied clinical and radiographic features imposing the fact that AOT should be included in differential diagnosis of routine odontogenic cysts and tumours.

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1. Introduction

Adenomatoid odontogenic tumour (AOT), called as the master of disguise, is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland in 1905. (Steensland, 1905) It is a hamartomatous, slowly progressive growth that accounts for 2.2–13% of all odontogenic tumours. (Batra et al., 2005) Philipsen and Birn proposed the name AOT, which was widely accepted and adopted by the World Health Organization classification of odontogenic tumors in 1971. (Philipsen and Birn, 1969) It is also known as ‘two-thirds tumor,’ because 2/3rd of cases occur in the maxilla, 2/3rd occur in young females, 2/3rd of the cases are associated with un-erupted teeth, and 2/3rd of the affected teeth are canines. (Garg et al., 2009) But this fascinating entity may camouflage as other odontogenic cysts/tumours, as seen in our three cases, two of which are atypical and one is a typical presentation of this tumour.

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2. Case 1

A 14 year old female patient presented with an incidental finding of a unilocular lesion in anterior mandible on the panoramic view which was taken in lieu of her orthodontic therapy. On clinical examination, mild labial expansion was palpable in mandibular incisor region. Increased interdental spacing was noted between left lower lateral incisor and canine (Fig. 1A). Panoramic view showed a 2X3 cms roughly circular well defined radiolucency surrounded by corticated borders extending from periapical region of right mandibular lateral incisor to left canine region. The lesion extended to the crest of interdental alveolar bone between left lateral incisor and canine. Distal displacement of roots of left mandibular canine was noticed. No root resorption was seen (Fig. 1B). 3D reconstructed CBCT view showed that the labial and lingual cortical plates were discontinuous and the unilocular lesion lied in close proximation to left inferior alveolar nerve canal at its exit in region of left mental foramen (Fig. 1C). Microscopy revealed duct like structures and cells proliferating in form of sheets (Fig. 1D).

3. Case 2

A 16 year old female complained of asymptomatic swelling on right side of chin since six months with insidious onset. She presented with a hard, non tender, non fluctuant swelling in right symphyseal region crossing midline. Obliteration of lower right anterior vestibule was seen. Over retained mandibular right and left canines were noticed with absence of its successor tooth(Fig. 1A). Panoramic view revealed a well defined, circular radiolucency with corticated borders which seemed attached to CEJ of impacted left canine. Right canine was displaced towards the inferior cortex which exhibited endosteal resorption. Root resorption was absent (Fig. 1B). CT scan showed buccal and mild lingual expansion associated with breaks in continuity of both cortical plates(Fig. 1C). Incisional biopsy showed spindle shaped epithelial cells proliferating as sheets and whorled rosette structures (Fig. 2D).

4. Case 3

A 17 year old male patient complained of swelling since 6 months following trauma and avulsion of left lateral incisor. A firm, non tender swelling was noticed in left anterior region obliterating the vestibule. Left canine was mesially drifted.

Maxillary occlusal view revealed a well defined corticated radiolucent lesion with small specks of calcification within the soft tissue delineations in relation with left canine (Fig. 3A). Solid capsulated mass (Fig. 3 B) revealed duct like stroma with lumina lined by single layer of columnar and cuboidal cells. (Fig. 3C).

5. Discussion

Adenomatoid odontogenic tumor is a benign, non-invasive lesion comprising of approximately 3% of all odontogenic tumors ranking behind odontomes, cementoblastomas, myxomas and ameloblastomas. (Kumar et al., 2010) AOT, as seen in our cases, is mostly encountered in the second decade of life. Our cases show female: male ratio of 2:1 which is consistent with the reported ratio of 1.9:1. (Garg et al., 2009) Most of the cases reported were in the maxillary canine region (Khot and Pavitra, 2011), but two of our cases did not follow this biological trend. The origin of AOT is controversial, but as it arises in the tooth bearing area, it is thought to arise from the odontogenic epithelium. (Yilmaz et al., 2009) Considerable amount of debate is still going on whether to consider AOT as a hamartoma or neoplasm. It may occur intraosseously and extraosseously. The intraosseous variety is of two types, most common follicular type associated with an unerupted tooth and a less common extrafollicular type which is not related to an impacted tooth. All of our cases are of intraosseous type with one follicular variant and two extra follicular variants.

Bicortical expansion was seen in cases 2 & 3 with resorption of cortical plates in all cases. Radiographically, it has unilocular radiolucency but multilocular variant are also reported which gives credence to the occurrence of multiple AOT. (Tsaknis et al., 1977) Minute radiopacities (califications) around the retained tooth considered characteristic, but not pathognomonic, were apparent only in one of our case. (Toida et al., 1990) Irregular root resorption is rare. This appearance must be differentiated from various types of disease, such as calcifying odontogenic cyst or tumor, dentigerous cyst, odontogenic keratocyst, ameloblastoma or periapical disease. (Kramer et al., 1992) Intra oral periapical radiographs allow better perception of even minimal calcifications in AOT. In addition, MRI has also been reported to be helpful in distinguishing AOT. The patients we described presented no root resorption, but displacement of the adjacent teeth, and also the tumor was not associated with an impacted tooth in two of the cases. (Toida et al., 1990) Radiographically, first case resembled odontogenic keratocyst and the second case resembled odontogenic keratocyst and the second case.

![Fig. 1](image-url)  A. Clinical image showing mild bicortical expansion in mandibular anterior region. B. Cropped panoramic image showing well-defined, unilocular, corticated radiolucent lesion in anterior region. Also, distal displacement of root of left canine is evident. C. CBCT showing loss of buccal and lingual cortical plates and approximation of lesion with IAN at its exit from mental foramen. D. Photomicrograph showing spindle and cuboidal shaped cells proliferating in the form of sheets. Typical duct like structures lined by a single layer of cuboidal to columnar epithelial cells can also be seen; these duct like lumina are lined by an eosinophilic rim (H & E, 10X).
resembled dentigerous cyst associated with impacted lower left canine tooth as it appeared attached to CEJ of the same. The typical location and presence of calcification flecks suggested AOT in 3rd case.

The histological features have been described as a tumor of odontogenic epithelium with duct like structures and with varying degree of inductive changes in the connective tissue. It may be partly cystic/ solid lesion may be present only as masses in wall of a large cyst. (Kramer et al., 1992) Our cases were consistent with these common features. The tumor is well encapsulated, therefore, conservative surgical enucleation produces excellent outcome without recurrence.

6. Conclusion

Our cases proffer insight into varied clinical and radiographic manifestations of AOT. It highlights the overlapping radiographic presentations, which an astute diagnostician should be aware of, while distinguishing more common lesions of odontogenic origin in routine dental examinations.

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