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Case Report

Two third's tumour – A case report

Varsha Patel G B [1,*], Sangeeth Siddabasappa[1], Ranjani Shetty[1], Ashok L[1]

[1] *Dept. of Oral Medicine and Radiology, Bapuji Dental College and Hospital, Davangere, Karnataka, India*

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ABSTRACT

Adenomatoid odontogenic tumour (AOT) is a rare, uncommon, benign odontogenic tumour that constitutes approximately 3% of all odontogenic tumors. It usually occurs in the second or third decade of life with female prediction. AOT commonly occurs in anterior maxilla. An accurate diagnosis should be established through clinical, radiographical and pathological correlations to differentiate AOT from other conditions. In this article we report a case of follicular AOT in 13-year-old female patient with swelling in left maxillary anterior region.

1. Introduction

Adenomatoid odontogenic tumour (AOT) is a rare odontogenic tumour that is frequently associated with unerupted teeth or dentigerous cysts, and is found in the anterior part of the maxilla without causing discomfort.[1] Adenomatoid odontogenic tumour, was originally described in the literature by Steensland in 1905. Philipsen and Birn proposed terminology for Adenomatoid odontogenic tumour with its simple abbreviation AOT and is now most widely accepted.[2,3]

AOT is also referred as the "two-thirds tumour" since two-thirds of the occurrences occur in the anterior maxilla in women and are linked with an included tooth, of which two-thirds are canines and two-thirds are intraosseous. Although tooth resorption is uncommon, cortical growth and adjacent tooth displacement are common.[4]

Based on clinical and radiographic findings AOT can be divided into 3 types – Follicular, extrafollicular and peripheral. Follicular is associated with crown of the embedded tooth, extrafollicular is not associated with embedded tooth and peripheral occurs on the gingiva.[5]

Radiographically the tumour presents as a well-circumscribed, unilocular radiolucency that is often associated with an unerupted tooth, most commonly a canine.

The purpose of this article is to report a case of AOT with an emphasis on clinical and radiographic features to enable proper diagnosis and treatment.

2. Case Presentation

A 13-year-old female patient came with the complaint of painless swelling on left anterior region of face since 1 month. Patient noticed the swelling 1-month ago which was insidious in onset, was initially smaller in size and gradually increased to the present size. The medical, dental and family history were non-contributory.

Extra oral examination revealed gross facial asymmetry with diffuse swelling present on left middle $1/3^{rd}$ of face, measuring about 2X3cm, extending mediolaterally from the ala of the nose to the zygoma and supero inferiorly from 2cm below the infra orbital margin to line joining the retrocommisure region to ear lobe, roughly oval in shape,

* *Corresponding author*.
E-mail address: varsha1996jan@gmail.com (V. Patel G B).

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skin and the surface over the swelling was normal, with obliteration of nasolabial fold and was not associated with any other secondary signs and symptoms. On palpation, there was no rise in local temperature, was non-tender, hard in consistency, non-compressible, non-fluctuant and non-mobile.(Figure 1)

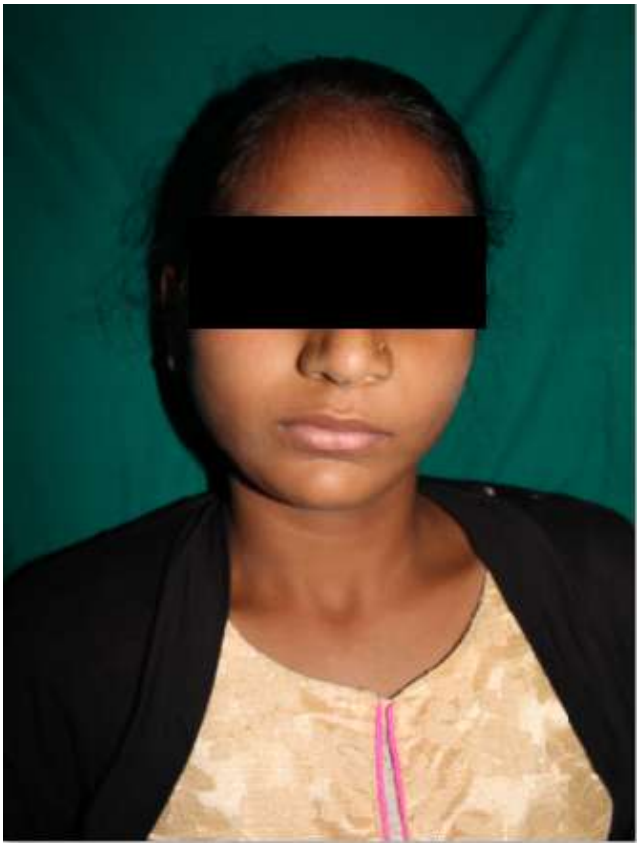

Fig. 1:

Right and Left submandibular lymph nodes were palpable, enlarged, measuring about 1x1cm in size, roughly oval in shape, non-tender, mobile, soft to firm in consistency.

Intra oral examination (Figure 2) revealed a solitary swelling in relation to left anterior maxillary vestibule, measuring 2x4cm in greatest diameter, extending mediolaterally from the distal surface of the left central incisor to the mesial surface of maxillary second premolar and superoinferiorly from the vestibule to the attached gingiva. The mucosa over the swelling was stretched and shiny. On palpation, swelling was non tender, bony hard in consistency with buccal cortical plate expansion. On hard tissue examination there was missing left permanent canine with over retained deciduous canine. (Figure 3)

Based on history and clinical findings a provisional diagnosis of benign odontogenic lesion in left anterior maxilla and a differential diagnosis of dentigerous cyst, adenomatoid odontogenic tumor and calcifying odontogenic cyst was considered.

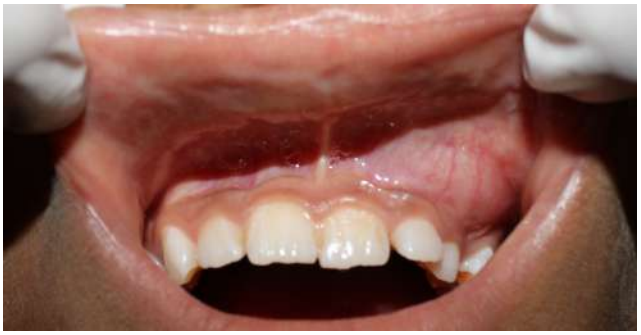

Fig. 2:

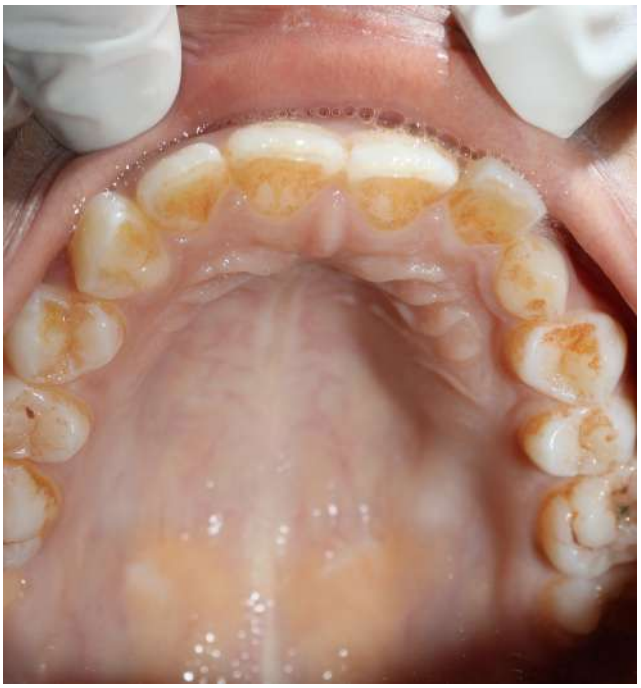

Fig. 3:

Orthopantomogram revealed a solitary well defined unilocular radiolucency with distinct sclerotic border and internal opacities in the left maxilla, extending mediolaterally from distal aspect of the root of left maxillary central incisors to mesiobuccal root of first molar with embedded permanent canine within the radiolucency. Root resorption was noted in relation to retained deciduous canine and the root of left lateral incisor was displaced medially and root of left first pre molar was displaced distally. (Figure 4)

The computed tomography taken in coronal section demonstrated the presence of well-defined cystic lesion in the left inframaxillary region measuring around 25 x 26 x 24 mm. There was an unerupted displaced tooth within the cystic lesion. The lesion was abutting the inferior portion of the left maxillary sinus and the lower portion of the left half

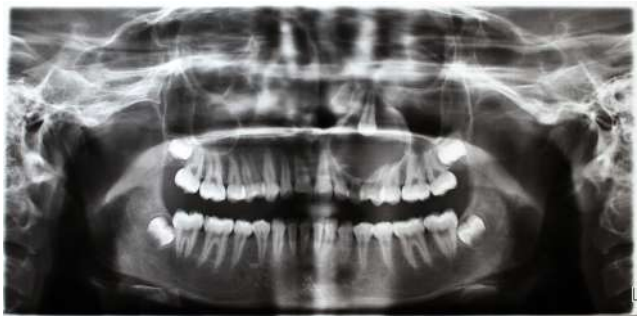

Fig. 4:

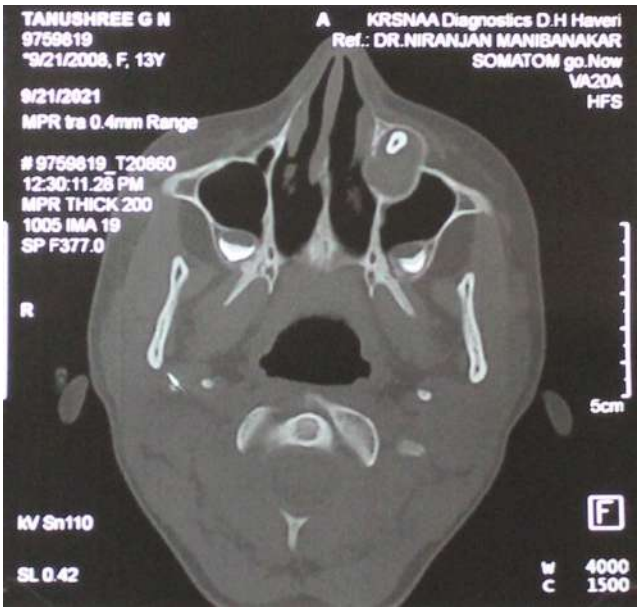

Fig. 5:

of nasal cavity. Nasal septum showed mild deviation to right side with bony spur.

Fine needle aspiration cytology was done and 3ml of strawed colored fluid was withdrawn from the lesion. It revealed the presence of numerous acute and chronic inflammatory cells and extravasated RBC's with very few exfoliated epithelial cells. Protein estimation of cystic fluid showed- 4.40 g/dl.

Surgical excision of the lesion was done. Excisional biopsy report revealed the presence of well circumscribed cystic connective tissue wall with central proliferation of duct like epithelium with small foci of calcifications. The epithelium appeared in rosettes double layer ameloblast like cells pattern, with eosinophilic fibrillar material present between tumor cells and within duct like structure. The tumor cells were tall columnar with basal nuclei and clear cytoplasm resembling pre ameloblasts. There was also presence of bony trabeculae with thick connective tissue wall which was highly cellular in nature. Few areas consisted of dilated blood capillaries and hemorrhagic space with extravasated RBC's. Correlating with the clinical and radiographic features, the above histopathological features were suggestive of adenomatoid odontogenic tumor.

3. Discussion

AOT is a noninvasive, odontogenic epithelial benign lesion which is a distinct entity in the maxillofacial region affecting young patients similar to our case. The latest WHO definition states "AOT is composed of odontogenic epithelium in a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth."[4] Adenoameloblastoma, ameloblastic adenomatoid tumor, epithelioma adamantinum, and teratomatous odontoma were all formerly used to describe the lesion now known as AOT.[1]

The origins of AOT are debatable. A number of theories have been advanced to explain the pathophysiology of AOT. It might possibly emerge from the enamel organ, the dentigerous cyst's epithelial lining, Malassez epithelial rests of the deciduous or permanent tooth or dental lamina remnants, and have an ameloblastic phenotype.[5]

AOT has distinct clinical features that make the diagnosis relatively obvious. First, this tumour is largely limited to young patients commonly between 3-28 years with an average age of 13 years. Second, its occurrence is confined to the anterior maxilla. Third, there is a female predominance with the ratio of 2:1. Our case reflects all the three features. Swelling are typically asymptomatic and hard in consistency. Involved teeth are usually impacted and adjacent teeth may be slightly displaced as in our case.[6]

The most frequently occurring pericoronal radiolucency in the jaw is the dentigerous cyst but it encloses only the coronal portion of an impacted tooth. In contrast AOT surrounds both coronal and radicular aspects of the involved tooth which resemble follicular variant of AOT similar to our case.[3]

Follicular AOT shows a well-defined unilocular radiolucency associated with the crown and often part of the root of an unerupted tooth. Extrafollicular AOT has no connection to an unerupted tooth and appear as a well-defined, unilocular radiolucency between, above, or superimposed upon the roots of erupted, permanent teeth. In the peripheral variant show some erosion of the adjacent cortical bone. Calcified deposits are seen in 78% of AOT.[3,7,8]

The histopathological features of AOT are very specific and all variants of AOT reveal similar histopathological characteristics. The lesion is lined by odontogenic epithelium enclosed within a capsule. Polyhedral and cuboidal epithelial cells within a fibro-collagenous stroma arranged in the classical "rosette" pattern were identified along with the presence of eosinophilic material with small

foci of calcified material which is similar to our case.[6]

Immunohistochemically, the classical AOT phenotype is characterized by a cytokeratin (CK) prole similar to follicular cyst and/or oral or gingival epithelium based on positive staining with CK5, CK17 and CK19 [11-13]. On the other hand the classical AOT is negative for CK4, 10, 13 and 18. Recently, Crivelini et al.[9] detected the expression of cytokeratin 14 in AOT and concluded that this probably indicate its origin in the reduced dental epithelium which is also positive for staining with cytokeratin 14 antibodies.

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Immunohistologically, AOT is characterised by cytokeratin (CK) profile similar to follicular cyst and/or oral or gingival epithelium based on positive staining with CK5, CK17 and CK19 but is negative for CK4, CK10, CK13 and CK18. Crivelini et al. detected the expression of cytokeratin 14 in AOT and concluded that this probably indicate its origin in the reduced dental epithelium which is also positive for staining with cytokeratin 14 antibodies.[10]

Enucleation and curettage are the most typical treatment options for the tumour.[3] AOTs are best treated with conservative surgical excision because of the low reported recurrence in the literature.

The above-described case is under follow up since 6 months after surgery with no recurrence.

4. Conclusion

The case reported here has the salient features of AOT described in the literature such as age, gender, anatomic region (anterior maxilla), unerupted teeth (canine), slow growing and unilocular radiolucency with calcification. It should be emphasized that although AOT is very rare, proper diagnosis and adequate integration of clinical and radiographic findings helps in arriving at correct diagnosis that helps in proper surgical management.

5. Source of Funding

None.

6. Conflicts of Interest

No conflicts of interest.

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Author biography

Varsha Patel G B, PG Student https://orcid.org/0000-0001-7636-3682

Sangeeth Siddabasappa, Professor

Ranjani Shetty, Professor

Ashok L, Professor and HOD

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