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Case Study

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ADENOMATOID ODONTOGENIC TUMOR: A CASE REPORT IN POSTERIOR MANDIBLE WITH A BRIEF REVIEW

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is a distinct odontogenic tumor which accounts for about 3-7% of all odontogenic tumours. It is a benign, non-invasive lesion, a painless swelling that is slow growing and gradually enlarging. AOT is predominantly found in females and usually arises in the second or third decade of life. It occurs in the maxilla for two third of cases. The occurrence is more common in young patients and two third of the cases is associated with missing or unerupted teeth. Treatment is conservative surgical excision and the prognosis is excellent. The present case is a rare report of an AOT presented in mandible of a 15-year-old female in association with impacted first premolar.

INTRODUCTION

Adenomatoid odontogenic tumour (AOT) is an uncommon benign odontogenic tumor and originates from the enamel organ epithelium. There are variety of terminologies that have been given to this entity such as adenoameloblastoma, adamantinoma, ameloblastic adenomatoid tumor, ameloblastic adenomatoid tumor or epithelioma adamantinum. It was first described by Dreibaldt in 1907 as pseudoadenoameloblastoma.[1] In 1969, Philipsen and Birn coined the term adenomatoid odontogenic tumour. It is also known as 'two-thirds tumor,' as 2/^{3rd} of the total cases are seen in in young females, 2/^{3rd} of cases are seen in the maxilla, 2/3rd are associated with unerupted teeth, and 2/^{3rd} of the cases are seen with the canines.[2] It is frequently found in young patients, especially in the second decade of life, affecting more commonly females in the ratio of male: female is 1:1.9.[1]

The WHO histological typing of odontogenic tumors, jaw cyst and allied lesions (2005) has defined AOT as a tumour of odontogenic epithelium with duct-like structures and with varying degree of inductive changes in the connective tissue.[3] There are three types of AOT that have described radiographically – Follicular, Extrafollicular and the Peripheral.[2] The follicular type (73%) constitutes of majority of cases followed by extrafollicular type (24%) and peripheral type (3%) that is seen the least of all and affects the gingival mucosa.[4]

CASE REPORT

A 15-year-old female patient reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling in the lower right back region of the jaw since past 1 month. History of the present illness revealed that initially the swelling increase in size gradually in the duration of 1 month. It was associated with dull localised, intermittent pain without any history of suppuration. Medical, dental and personal histories were not relevant. On extraoral examination, no contributory result was obtained.

Intraoral examination revealed an ovoid, soft, fluctuant swelling present on the right posterior mandibular region extending from 43 to 45 region with obliteration buccal vestibule. The size of the swelling is approximately about 2cm×3cm and colour of overlying mucosa was

same as that of contralateral region. There were no lymph nodes palpable in association with the lesion. On palpation, the lesion was soft in consistency. On hard tissue examination, displacement of 43, 45 with clinically missing 44 was observed.

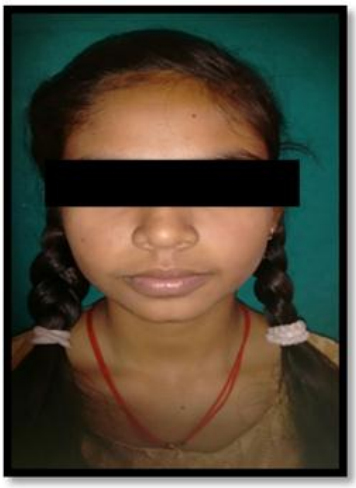

Figure 1: Extra oral examination.

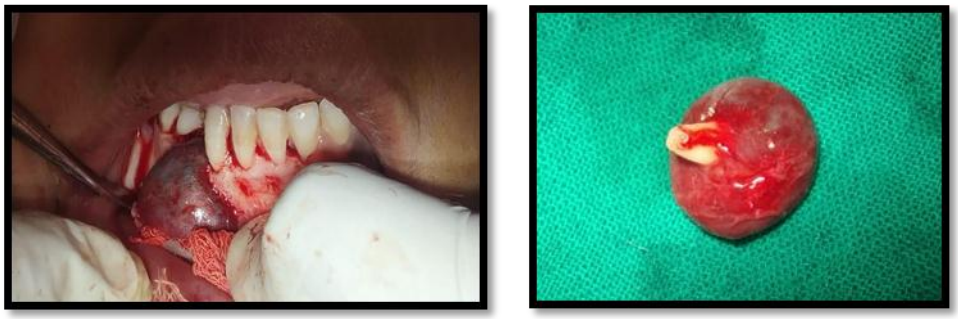

Figure 2: Intraoral examination.

Figure 3: Fine needle aspiration cytology revealed a straw-coloured fluid.

An OPG revealed a well circumscribed radiolucency with intact sclerotic borders is seen wrt. 43,45. Extending from the alveolar ridge to the inferior border of the mandible superioinferiorly and from the distal aspect of 43 to the mesial aspect of 46 mesiodistally. The radiolucency is seen encircling the impacted 44. The radiolucency is seen pushing 43 distally and 45 mesially without any root resorption.

Haematological evaluation showed normal parameters. An excisional biopsy was performed under local anaesthesia and the tissue sample was submitted for histopathological evaluation to the Department of Oral Pathology and Microbiology, Santosh Dental College and Hospital.

Considering the patient history, clinical examination and radiographic examination a provisional diagnosis of dentigerous cyst was made.

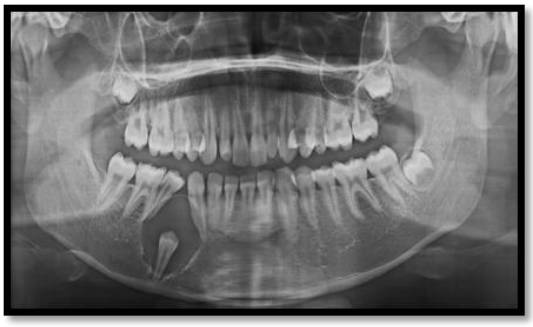

Figure 4: Orthopantogram.

Histopathological examination

All the bits of soft tissue specimen obtained from the patient were grossed. The specimen was black-brown in colour, firm in consistency & ovoid in shape which measured about 2 x 3cm (Figure 5).

Figure 5: Grossing.

Microscopically the section shows multinodular proliferation of neoplastic cells. The neoplastic cells are spindle, cuboidal and columnar in shape. At some places duct like structures are noted that are lined by a single layer of cuboidal or columnar cells with nuclei polarised away from the lumen. Few areas show presence of rosette like structure with a central eosinophilic material. Scattered foci of calcification can also be appreciated with scanty connective tissue stroma.

Overall features are suggestive of Adenomatoid Odontogenic Tumor (figure: 6-8)

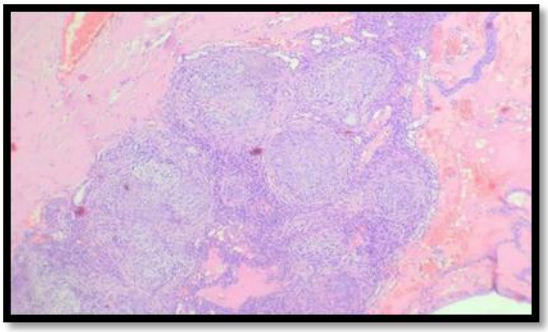

Figure 6: 4x: The section shows multiple nodules of epithelium and the connective tissue stroma.

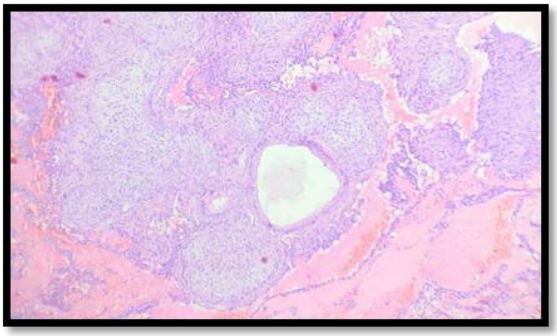

Figure 7: 10x: The duct like spaces is lined by a single row of columnar cell and epithelial cells forms rosette like pattern.

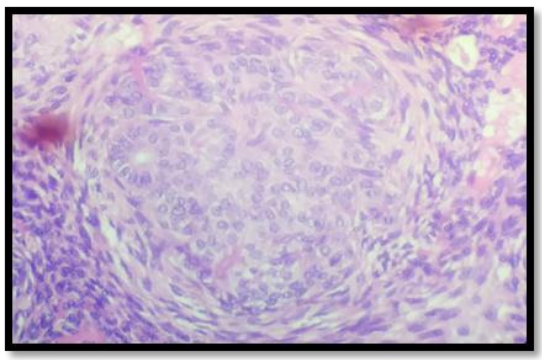

Figure 8: 40x Odontogenic epithelial cells arranged in rosette pattern.

DISCUSSION

Adenomatoid odontogenic tumor (AOT) is slow growing, benign tumor of odontogenic origin and is relatively rare.[5] In 1969, the Philipsen and Birn coined the term "adenomatoid odontogenic tumor" by which it is popularly known. In 1971, this terminology was included in World Health Organization (WHO) histological typing of odontogenic tumors, jaw cysts and allied lesions. AOT was redefined in 2005 by WHO as a tumor that is composed of odontogenic epithelium, presenting with a variety of histoarchitectural patterns, embedded in mature connective tissue stroma and characterized by progressive slow growth.[6]

Histiogenesis

The origin of AOT is still controversial. Most of the authors believe it to be odontogenic in origin as it occurs within the tooth bearing areas of the jaws. Whereas, some studies proposed that the cytological features of AOT is similar to enamel organ, dental lamina and reduced enamel epithelium. It is also said that the lesion is developmental outgrowth or hamartoma due to the relative size of the tumor and lack of recurrence.[5]

Clinical Features

It is prevalent in 2nd to 4th decade of life especially in the females. It is an asymptomatic, benign, slow growing tumor, which is predominately seen in anterior maxilla, associated with incisors, canine and 1st premolars.[6] The teeth involved are generally impacted and the adjacent teeth tend to get displaced. Basically, the tumours do not exceed 1–3 cm in greatest diameter but they can be larger. They usually occur within the dentate areas of jaws and often found usually with impacted or unerupted teeth.

Brief Review

The present case of AOT presents its rarity in the form of unusual location of occurrence i.e., right mandibular posterior region. The usual occurrence of AOT was confined mostly to maxillary unerupted canine. After extensive research on the literature, we retrieved few cases reports in which the AOT was found in the posterior mandibular region (Table-1).

Table -1.

S. No	Author's	Gender	Age	Site
1	Ming-Jane Lang et al.[7] (2015)	F	25yrs	Posterior mandible- molars
2	Gomez et al.[8] (2013)	M	32yrs	Posterior mandible- molars
3	Miles et al.[9] (1951)	M	18yrs	Posterior mandible- molars
4	Allen et al.[10] (1998)	M	37yrs	Posterior mandible
5	Belgaumi, et al.[11] (2015)	M	21yrs	Mandibular Premolar
6	Kemp et al.[12] (2008)	M	46yrs	Right mandibular body and ramus
7	Bhullar RP at al.[13] (2011)	F	24yrs	Mandibular Premolar
8	Martínez et al.[14] (2009)	F	10yrs	Mandibular Molar
9	Shivali V et al.[15] (2013)	M	18yrs	Mandibular Molar
10	Allen et al.[10] (1998)	M	35yrs	Left posterior mandible

The literature showed that there are very few cases of AOT reported in posterior mandibular region. Amongst majority of published reports, AOTs are associated to molar regions having male predilection. The last case reported of AOT in posterior mandibular region is in 2015. The current case report presents the lesion associated with young female in association with impacted first premolar, which makes this case unique from the others.

Variants of AOT

Radiographically, all cases present as well-defined unilocular radiolucency with corticated borders and associated with impacted teeth. The AOT have three clinicopathological variants: Follicular, Extrafollicular and Peripheral. The follicular and extrafollicular variants account for almost 97.7% of all AOTs.[16] They are generally present as intrabony tumors. Peripheral AOT (PAOT) presents as a gingival growth with a significant predominance in females and is seen in the anterior maxilla, and it is primarily involved with the incisors. All the findings of AOT frequently resemble other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, calcifying globule-maxillary cysts, odontogenic tumors, ameloblastomas, odontogenic keratocysts and periapical pathologies.[17]

Histopathological Features

AOT microscopically presents a range of unique and distinctive features such as - occurrence of varying size nodules of cuboidal or columnar epithelial cells forming nest or rosette like structures in the background of scanty connective tissue stroma. Amorphous homogeneous material that is eosinophilic is usually seen in the core of these rosettes. They can form duct-like spaces of varying diameter, but this pattern is not frequently seen.[18] The ducts are lined by a single layer of cuboidal or columnar epithelial cells that have nuclei with reverse polarisation. Darkly stained dystrophic calcifications in minute amounts are also observed in the histological sections.

Immunhistological features

During the last few years several studies have been published dealing with the immunhistological properties of AOT. Immunohistochemically, the classical AOT phenotype is characterized by a cytokeratin (CK) profile similar to follicular cyst and/or oral or gingival epithelium based on positive staining with CK5, CK17 and CK19.[19] On the other hand, the classical AOT is negative for CK4, 10, 13 and 18. Recently, 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. Positive reactions for amelogenin in limited areas in AOT are also reported as well as in ameloblasts and in the immature enamel matrix.[20]

CONCLUSION

In the present case the site of occurrence AOT is rare. In the reviewed literature, till now only 10-15 cases were reported in the region of posterior mandible. These cases were mostly commonly found in the molar region with a male predilection. In present case report, the lesion is associated with 1st premolar and in a young female. Therefore, thorough histopathological evaluation is required of the excised lesion that will contribute to the accurate definitive diagnosis and appropriate management. AOT must be considered as a differential diagnosis of the swellings until all the other possibilities are histopathologically ruled out.

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