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Case Report

Adenomatoid Odontogenic Tumor - The "Two-thirds tumor"

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ABSTRACT

Adenomatoid odontogenic tumor is an uncommon benign hamartomatous lesion of odontogenic origin, commonly seen in adolescents with a female predilection. It has distinct features such as constrained size, minimal growth potential and lack of recurrence. It occurs in both intra-osseous and peripheral forms. The rarity of adenomatoid odontogenic tumor may be associated with its asymptomatic presentation and slow growing pattern. Therefore, it should be distinguished from the more common lesions of odontogenic origin in routine dental examinations. Here we report a case of adenomatoid odontogenic tumor in a 14 year old female patient in the anterior maxillary region. The patient was treated surgically and later rehabilitated with fixed prosthesis.

KEYWORDS: Adenomatoid odontogenic tumor; Hamartoma; Impacted tooth; Maxilla.

INTRODUCTION

Adenomatoid odontogenic tumor, is an uncommon benign epithelial lesion of odontogenic origin accounting for 2.2% to 7.1% of odontogenic tumors as reported in a recent study [1]. Adenomatoid odontogenic tumor was first described by Dreibladt, in 1907, as a pseudoadenoameloblastoma [2].Stafne in 1948 considered it a distinct entity, but was classified by others as a variant of ameloblastoma [3]. Later Philipsen and Birn proposed the name adenomatoid odontogenic tumour in 1969 and suggested that it should not be regarded as a variant of ameloblastoma because of its different biological behavior [4]. This term was adopted by the World Health Organization (WHO) classification in 1971 [5]. WHO classification (2005) defines AOT as "being composed of odontogenic epithelium in a variety of histo-architectural patterns, embedded in a mature connective tissue stroma characterized by slow but progressive growth."

There are 3 variants of adenomatoid odontogenic tumour - the Follicular type (accounting for 73% of cases) is a central intrabony lesion associated with an unerupted tooth; the extra follicular type (24% of case) is also an intra-osseous lesion but unrelated to an unerupted tooth and the peripheral type is a rare variety (3% of cases) that arises in gingival tissue [6-9]. The tumor appears as an intraoral-extraoral swelling in the maxilla, and is sometimes referred to as "Two-thirds tumor" because it occurs in the maxilla in about 2/3 cases, about 2/3 cases arise in young females, 2/3 cases are associated with an unerupted tooth, and 2/3 affected teeth are canines [10]. The increasing number of reports on AOT points to the fact that the tumor develops more

frequently than formerly expected. This report includes a further depiction of AOT with particular reference to the clinical and diagnostic (radiological, histological and immunohistochemical) feature.

CASE REPORT

A 14 year old female patient reported with a chief complaint of swelling on the right side of face and mild discomfort since 20 days. The medical and dental history was insignificant and patient was in good general health. There was no history of facial trauma. Extra oral examination revealed a small swelling with the obliteration of the nasolabial fold (Fig. 1). Intraoral examination revealed an intra-osseous lesion of the right maxilla causing expansion of the buccal cortical plate, obliterating the labial vestibule. There was disruption of the normal orientation of the anterior teeth. The overlying oral mucosa was apparently unaffected (Fig. 2). On palpation the swelling was bony hard, 2.0 × 2.5 cm in diameter with a well‑defined border. Hard tissue status revealed missing 11 and over retained 51.

Figure: 1 Extra-oral picture showing right maxillary swelling

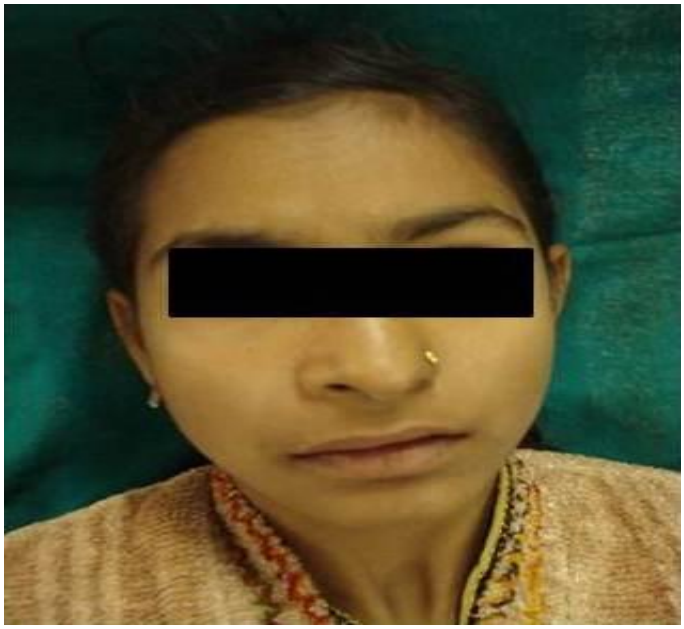
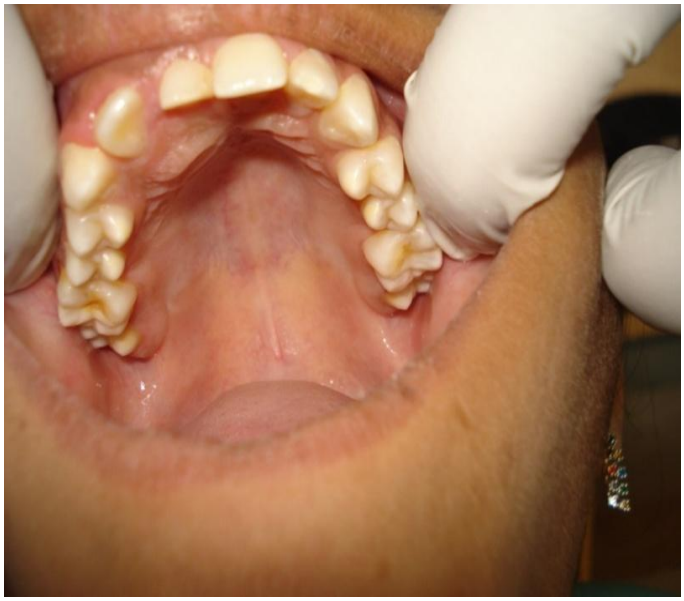


Figure: 2 Intraoral picture showing swelling of the right maxillary palatal region with missing right permanent incisor, displaced right lateral incisor and canine and over-retained deciduous incisor.



Radiographs: Maxillary occlusal view revealed the presence of a significant unilocular radiolucent area with well-defined sclerotic borders involving an impacted upper right permanent incisor, root resorption in relation to 51 and rotation of the right permanent lateral incisor (Fig. 3).

Fine Needle Aspiration: Straw colored fluid. Based on clinical and radiological findings, a provisional diagnosis of dentigerous cyst associated with impacted 11, with differential diagnosis of adenomatoid odontogenic tumor, keratocystic odontogenic tumor and unicystic ameloblastoma was made.

Under local anesthesia, excisional biopsy of the lesion was performed with excavation of upper right incisor. Grossly the lesion appeared as a globular mass measuring 2 × 2 × 2 cm in size, creamish brown in color, having a firm consistency and was attached to the cervical area of the tooth .

Histopathology (Fig. 4) revealed islands of odontogenic epithelium proliferating into connective tissue. The tumour islands predominantly consisted of polyhedral or spindle shaped cells, arranged as large islands or solid sheets, whorled pattern and some areas showed duct‑like pattern lined by cuboidal to columnar cells at the periphery. In some ducts, band of eosinophilic (amyloid-like) material lining the single layer of cells on the luminal side was seen. Some areas showed cells arranged in multiple layers and in between these layers, a band of eosinophilic material gave a typical rosette pattern to the tumor. A well defined fibrous tissue capsule was seen at the periphery and foci of extravasated red blood cells were seen in few areas. Correlating the clinical, radiological and histopathological features the diagnosis of Intraosseous Follicular Adenomatoid Odontogenic Tumor in association with impacted 11 was made.

Recovery was uneventful and the edentulous space was managed by use of a fixed partial denture. There was no recurrence for a follow-up period of two years.

Figure: 3 Occlusal radiograph showing radiolucency surrounding the impacted right permanent incisor, over-retained deciduous incisor and displaced lateral incisor and canine.

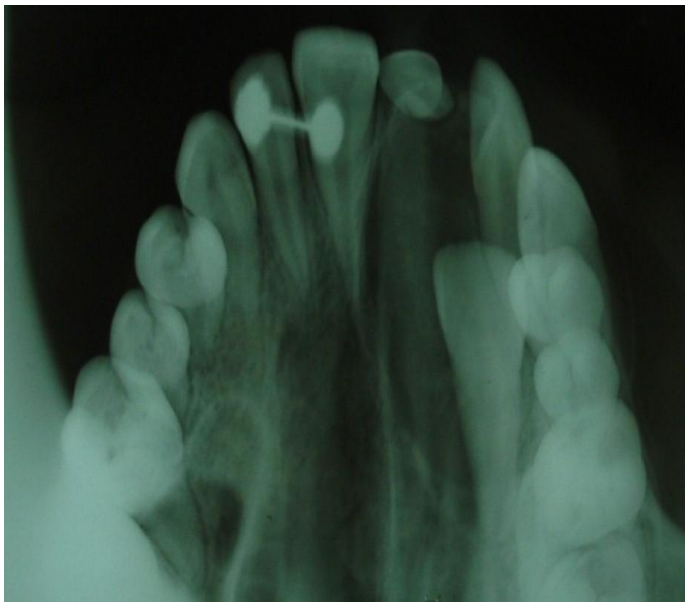
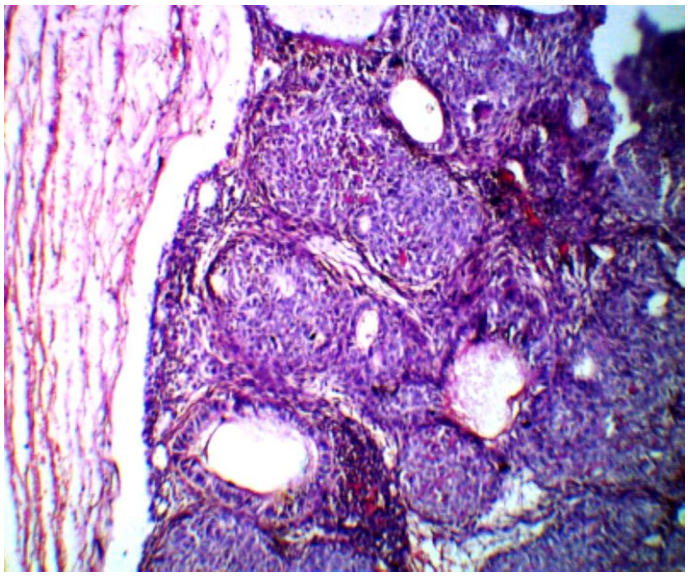


Figure:4 Histopathological section showing highly cellular tumor made up of nests and solid islands lined by cuboidal and low columnar cells arranged in whorls, ductular and ring-like patterns surrounded by an intact capsule(H& E stain x200).



DISCUSSION

Adenomatoid odontogenic tumour is a slowly growing lesion, commonly occurring in maxillary anterior region (ratio of cases 2:1 relative to mandible) of young females and is mostly diagnosed in the second decade of life, more than half occurring during the teenage years, as in the case reported here. The female to male ratio for all age groups and all variants is close to 2:1 [11]. The lesions are typically asymptomatic, commonly involving an impacted tooth and may be discovered during routine radiographic examination. It may appear as a painless hard swelling and can cause cortical expansion and displacement of the adjacent teeth, as in the case reported here [12]. Radiographically, the intraosseous follicular type of AOT shows a well-delineated, unilocular radiolucency surrounding the crown of an impacted tooth, a picture indistinguishable from follicular cysts. Indeed, AOT and other odontogenic lesions such as calcifying odontogenic cyst/tumor, ameloblastoma, dentigerous cyst, keratocystic odontogenic tumor or periapical disease share similar radiological features [13, 14].

The subtyping of AOT is based on clinical and radiological findings. The follicular (intraosseous) type is by far the most frequent growth type of AOT. The tumor in the case of follicular type surrounds the crown of an impacted tooth and may extend to cover the upper part of the root. The gross macroscopic appearance of the tumor was in accordance with former definitions of the entity recommended by the WHO[5] that AOT may be "partly cystic and in some cases the solid lesion may be present only as masses in the wall of a large cyst", as in this case. The tumor is thought to be a hamartoma rather than a true neoplasm [7], but currently there is no evidence to resolve this dispute. Recent reports indicate that the cells of an adenomatoid odontogenic tumor usually differentiate toward an apparent ameloblastic phenotype but fail to achieve further functional maturation [12].

According to WHO the histological findings for AOT are remarkably similar in the literature. The histological features of the tumor were described as a tumor of odontogenic epithelium with duct like structures and with varying degree 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 [15]. The tumor may contain pools of amyloid-like material and globular masses of calcified material [16]. The findings of our case were in accordance with the features reported in the literature. The histologic appearance of all variants is identical and exhibits remarkable consistency.

Immunohistochemistry of AOT expresses cytokeratin (CK) profile similar to follicular cyst and/or oral or gingival epithelium based on positive staining with CK5, CK17 and CK19 [17].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 [18]. Positive reactions for amelogenin and enamelin are seen in small mineralized foci of the tumor and in hyaline droplets in limited areas and these proteins are reported as well in ameloblasts and in the immature enamel matrix [19]. Interestingly, Takahashi et al. observed a positive staining for iron-binding proteins (transferring, ferritin) and proteinase inhibitor (alpha-one-antitrypsin) in various cells of AOT indicating their role in the pathogenesis of AOT [20].

Since all variants show identical benign biological behavior and almost all are encapsulated, curettage or conservative surgical enucleation is the treatment of choice. Encouragingly the prognosis is good and recurrence is very

rare after complete removal of the lesion and there have been no reports on aggressive behavior of AOT.

CONCLUSION

Adenomatoid odontogenic tumour is a slowly growing lesion, commonly occurring in maxillary anterior region of young females and is mostly diagnosed in the second decade of life during the teenage years. It occurs in the maxilla in about 2/3 cases, about 2/3 cases arise in young females, 2/3 cases are associated with an unerupted tooth and 2/3 affected teeth are canines and so it is also referred to as "Two-thirds tumor." The rarity of AOT may be associated with its slowly growing pattern and symptomless behavior. Therefore, it should be distinguished from more common lesions of odontogenic origin in routine dental examinations. The histologic appearance of all variants is identical and exhibits remarkable consistency. Curettage or conservative surgical enucleation is the treatment of choice, as all variants show identical benign biological behavior and almost all are encapsulated.

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