Case Report

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First Case Report of Adenomatoid Odontogenic Tumour Mimicking a Periodontal Intrabony Defect in a Young Male

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ABSTRACT

Background: Adenomatoid odontogenic tumour (AOT) is relatively a rare odontogenic tumour seen in younger age group, with site predilection for anterior maxilla exhibiting female predominance. This is a report of AOT mimicking a periodontal intrabony defect in a young male, which was managed through conservative surgical approach with positive follow up results. This paper also highlights various clinical manifestations, histological features and management aspects of AOT.

Keywords: Adenomatoid odontogenic tumour; case report; odontogenic tumours.

INTRODUCTION

AOT was denominated as "cystic adamantoma" by Harbitz in 1915, Stafne considered it as a distinct entity in 1948. Philipsen and Birn proposed the name adenomatoid odontogenic tumour in 1969 and suggested that it cannot be regarded as a variant of ameloblastoma because of its different behaviour[1]. This term was accepted by the WHO classification in 1971 [2]. The lesion was initially referred to as adenoameloblastoma, ameloblastic adenomatoid tumour, glandular ameloblastoma, adenomatoid ameloblastoma [3]. AOT comprises of only 0.1% tumours and cysts of jaws and constitutes about 3% of all odontogenic tumours [4]. The tumour is keen to involve the anterior maxilla in the lateral incisor and canine region with female bias [5]. AOT occurs as peripheral and central variants, the central being of follicular (with embedded tooth) and extrafollicular type (with non embedded tooth). Out of which the follicular variety is considered more common [6]. This article illustrates a case of AOT in the maxillary anterior region resembling a periodontal defect in a young male patient.

CASE REPORT

A sixteen-year-old male reported with displacement of maxillary left lateral incisor. There was no correlating history of pain, swelling or trauma. Intraoral examination revealed full complement of teeth except the third molars. The maxillary left lateral incisor was displaced labially, gingiva related to the tooth was normal in appearance with no evidence of expansion of the cortical plates. Deep defect was noted on the palatal aspect on periodontal probing (Fig. 1). Radiographic examination revealed a well defined homogenous radiolucency with regular corticated borders distal to maxillary left lateral incisor, extending apically, causing displacement of the teeth contralaterally with no evidence of root resorption (Fig. 2). The tumour was completely enucleated with thorough curettage of the region under local anesthesia by raising the palatal flap. Histopathological examination revealed proliferating odontogenic

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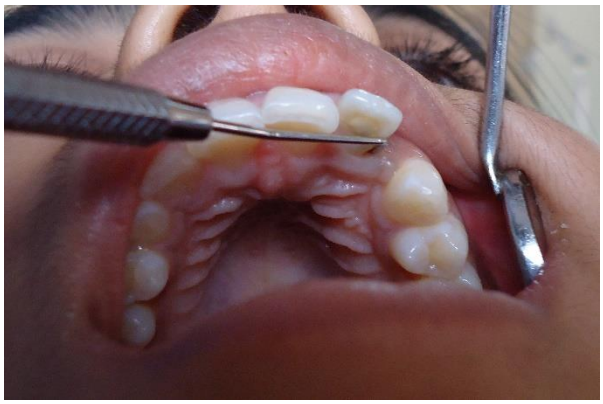

Fig 1: Deep palatal pocket.

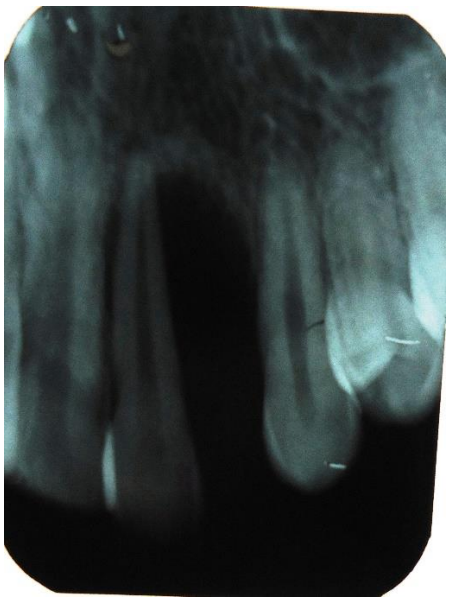

Fig 2: Preoperative intraoral radiograph showing well defined deep palatal defect.

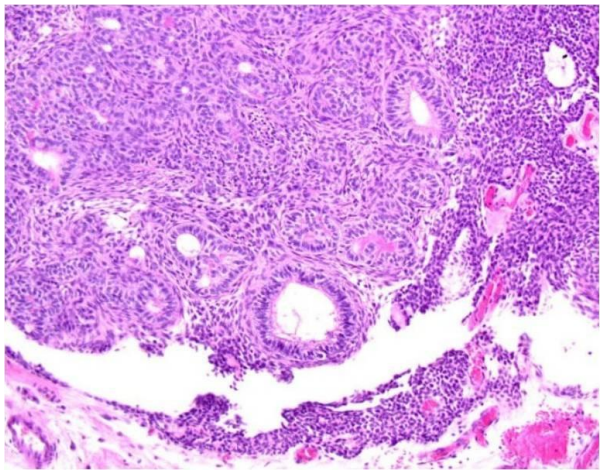

Fig 3: Photo micrograph (10x) showing proliferating odontogenic cells arranged in duct like Pattern, amyloid like material & fibrous capsule.

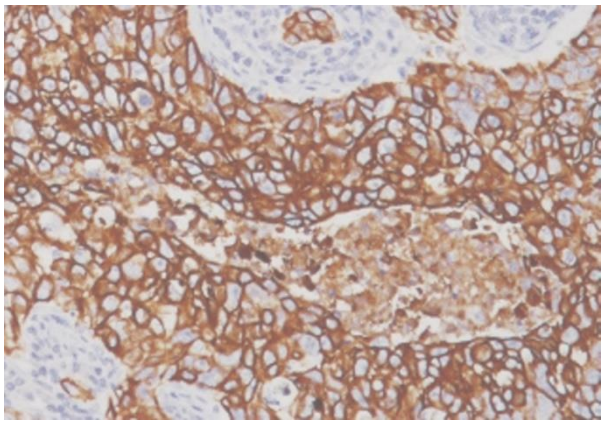

Fig 4: Immunohistochemical staining revealing whorled epithelial cells and glandular pattern.

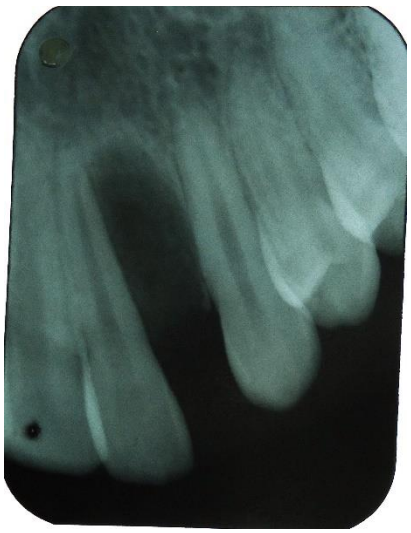

Fig 5: Six month postoperative radiograph showing healing lesion.

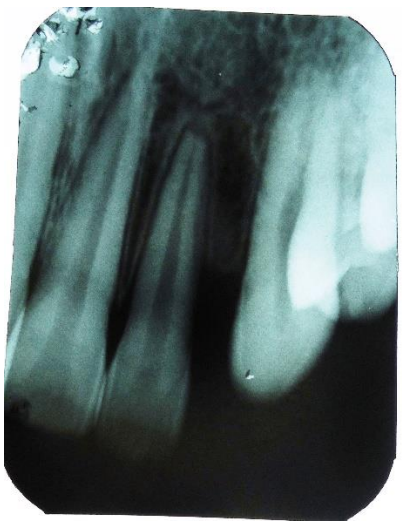

Fig 6: Intraoral periapical radiograph after twelve months.

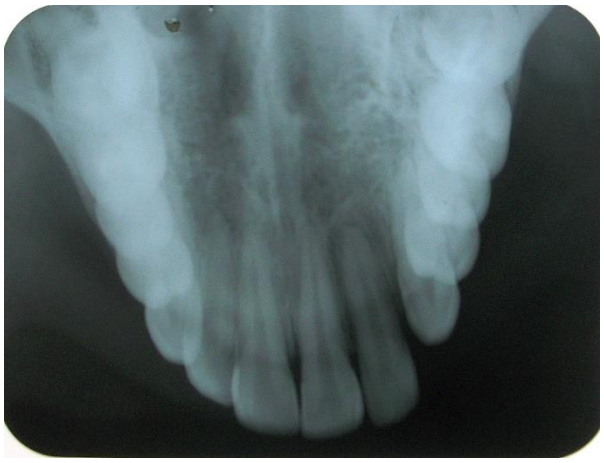

Fig 7: Occlusal radiograph showing uneventful healing.

cells arranged in duct like pattern, whorled masses and in sheets. Amorphous eosinophilic material is seen between the cells. The intervening connective tissue is scanty and shows foci of calcification. Fibrous connective tissue capsule and areas of haemorrhage are seen in the periphery (Fig. 3). Immunohistochemical analysis showed expression of keratin and vimentin at the periphery and superficial duct-like structures along with whorled epithelial cells and glandular pattern (Fig. 4). Follow up conducted after six months revealed a healing postoperative defect (Fig. 5) and twelve months showed uneventful healing with radiographic evidence of defect resolution, no signs of recurrence were noted (Fig. 6,7).

DISCUSSION

AOT is considered as rare cause of jaw swelling, affecting more commonly the females than males with the ratio being 2:1 [7]. In contrast to this, Chattopadhyay reported thirty cases of AOT from India in which he found male predominance [8]. AOT is more prevalent in young patients, especially during second decades [9]. The features as observed in this patient correlate with the data mentioned above. These tumours are usually asymptomatic, may cause cortical expansion, displacement of teeth and rarely root resorption [2]. Radiographically the lesion appears as a unilocular radiolucency, with mild radioopaque foci seen occasionally. The origin of AOT is controversial, although few researchers consider this entity as hamartomatous proliferation of odontogenic tissues [10]. Majority of the authors believe that AOT originates from odontogenic source, as it occurs within the tooth bearing areas in association with embedded teeth and having histological features similar to those of odontogenic components [1].

Similar and consistent histological features are seen in all the variants of AOT. It is described as a tumour of odontogenic epithelium with duct like structures and variable degree of inductive changes in connective tissue. The tumour is partly cystic, however in some cases the solid lesion may be present as masses in the wall of a large cyst. Pools of amyloid like material and globular masses of calcified material have been noted as components of AOT [5]. The features seen in this case were consistent with those reported in the literature. Immuno-histological studies of the lesion shows expression of the keratin and vimentin in the tumour cells at the periphery of the ductal, tubular or whorled structures along with small mineralized foci of amelogenin and enamelin found in the tumor cells and in hyaline droplets [2]. Encapsulation and benign biological behaviour point towards conservative surgical treatment or curettage as the treatment of choice, with minimal chances of recurrence [9]. Keeping these aspects in view, this was managed on similar backgrounds yielding remarkable results.

CONCLUSION

The report describes a case of extra follicular variant of AOT resembling periodontal bone defect are, which is hereby managed through conservative intervention with positive outcome. Rarely any such case is reported in literature till date.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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