Surgical management of peripheral variant of adenomatoid odontogenic tumor: A rare case report with review

Karuna Jindwani, Y. K. Paharia, Atul Pratap Singh Kushwah

Abstract

The adenomatoid odontogenic tumour (AOT) is a relatively uncommon lesion constituting around 3% of all odontogenic tumours and often misdiagnosed as an odontogenic cyst. It manifests as a beningn growth which affects young individuals, with a female predeliction usually in the second decade of life, exhibiting more often in the anterior region of maxilla. The current article enumerates the clinical, radiographic and histopathological features of a rare case of extraosseous AOT with its therapeutic consideration

Keywords: Adenomatoid odontogenic tumor, extraosseous, odontogenic cyst

Introduction

Adenomatoid odontogenic tumor (AOT) is a hamartomatous, slowly progressive growth that accounts for 2.2–13% of all odontogenic tumors.[1] It usually affects teenagers and occurs in the middle and anterior portions of the jaws. Approximately about two-thirds of these tumors occur in the maxilla and are more common in young females. The cases are associated with unerupted teeth and mostly maxillary canine.[2,3] It often causes the expansion of the surrounding bone and the displacement of the adjacent teeth.[4]

It was first described by Dreilbaldt in 1907 as a pseudo-adenoameloblastoma[5] and was first reported by Harbitz in 1915 as a cystic adamantinoma.[6] A variety of terms has been used to describe this tumor like adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma admentinum or teratmatous odontoma.[7] Philipsen and Birn proposed the widely accepted and currently used name AOT which was adopted by the World Health Organization classification of odontogenic tumors in 1971.[8]

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This case report highlights an unusual case of peripheral AOT, mimicking an odontogenic or eruption cyst originating in maxillary left anterior region of a 21-year-old male.

Case Report

A 21-year-old male presented to the Department of Dentistry, Government Medical College, Gwalior with a chief complaint of growth in his upper left front teeth region that had persisted for a year. The swelling had gradually increased in size with no associated pain, discharge or numbness.

Extra-oral physical examination revealed a single swelling in the left anterior maxillary region giving a local raised lip appearance [Figure 1]. Intra-oral examination revealed a solitary well-circumscribed firm growth measuring about 2 cm × 2 cm situated on the gingiva labial to 22 extending from distal aspect of 21 till mesial aspect of 23 causing palatal displacement of 22 and labial drifting and slight rotation of 21 mesially [Figure 2]. On palpation, the swelling was firm in consistency and showed no evidence of discharge on digital pressure. It was non tender with a smooth surface and was present labial to palatally displaced 22.

The patient was subjected for radiographic investigations: Intraoral peri-apical radiograph of the left maxillary anterior region, maxillary anterior occlusal view [Figure 3] and an orthopantomograph (OPG). The radiographs revealed a well-defined radiolucency measuring about 2 cm × 2 cm extending from the distal aspect of 21 to mesial aspect of 23 and confirmed displacement of 22.

Based on the clinical features, history and radiographic examination, a provisional diagnosis of a benign neoplasm/cyst of odontogenic or non-odontogenic origin was made.

The patient underwent surgical curettage (enucleation) and extraction of 22 under local anesthesia in outpatient department of our institute and the samples were sent for histopatologic examination.

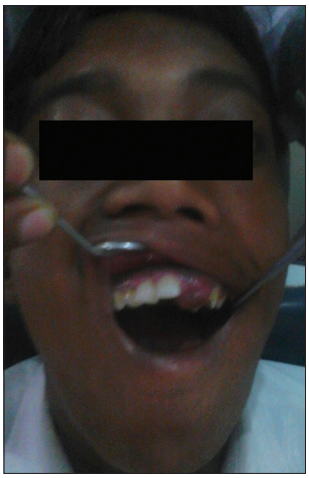

Figure 1: Extra-oral facial profile

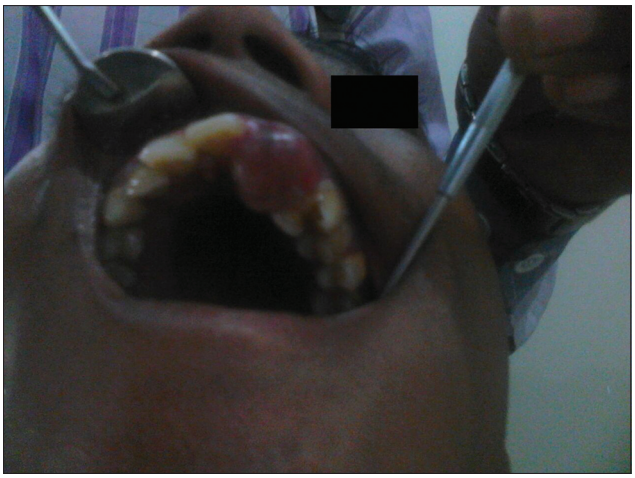

Figure 2: Intraoral view

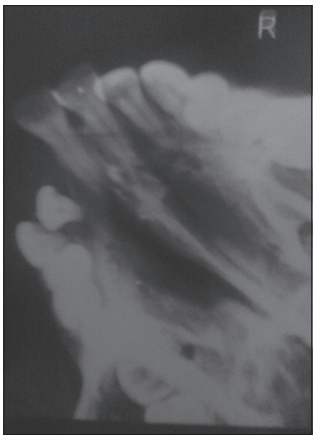

Figure 3: Occlusal view of the maxilla

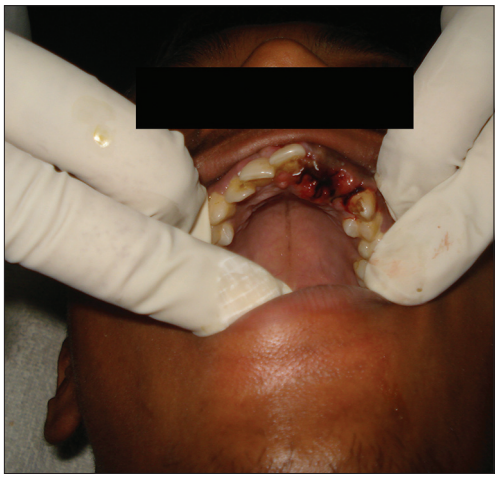

Figure 4: Intra-operative – Removal 22

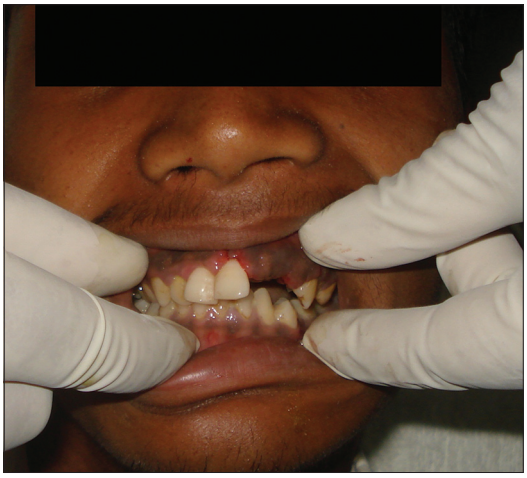

Figure 5: Post-operative

The tumor cavity was packed with gelfoam and closed with 3-0 black silk interrupted sutures [Figure 4]. Healing was uneventful, with no evidence of recurrence after surgery [Figure 5].

Microscopy revealed an odontogenic lesion characterized by proliferation of spindle-shaped cells, arranged as whorls, sheets and strands. Rings of columnar cells gave rise to duct like appearance. Connective tissue was scanty with areas showing large blood vessels. The overall features were suggestive of AOT. Thus, the final diagnosis of peripheral variant of AOT was obtained.

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.[9,10] AOT is mostly encountered in young patients, especially in the second decade of life. Females are affected more often than males with a female: Male ratio of 1.9:1.[2,3,11,12] The maxilla is the predominant site of occurrence, being almost twice as frequent as that in mandible. AOT present as a slow-growing symptom free lesion which more frequently involves the anterior part of the jaw. The origin of AOT is controversial, but as it arises in the tooth bearing area, it is thought to arise from the odontogenic epithelium.[13] The tumor has three clinic pathological variants, namely intraosseous follicular,

intraosseous extra follicular, and peripheral. The follicular type is a central intrabony lesion associated with an unerupted tooth, which accounts for about 73% of all the AOT cases. The extrafollicular type is also an intra-osseous lesion but not related with an impacted tooth, which accounts for about 24% of all the AOT cases and the peripheral variant is rare form and is attached to the gingival structures which accounts for about 3%.[14,15]

In our case, a rare peripheral variant of AOT occurred primarily in the palatal gingival tissue of the anterior tooth bearing area of maxilla on the left side in a young male which is in accordance with the literature.

A distinct radio-opaque border of the unilocular radiolucency is a characteristic radiographic manifestation of AOT. The extraosseous, peripheral or gingival type of AOP are rarely detected radiographically, but there may be slight erosion of the underlying alveolar bone cortex.[11] It usually manifests with the displacement of the teeth that was evident in our case on the OPG and occlusal views of the patient.

Histologically, AOT is highly unusual, as it presents a variety of cellular patterns. The characteristic duct-like structures are lined by a single row of columnar epithelial cells, the nuclei of which are polarized away from the central lumen. Dystrophic calcification is sometimes seen and can be extensive.[11] In our case, mixed picture with basophilic cells forming whorls seen at some places and at other sites columnar cells constituting duct-like structures confirmed it to be AOT.

Owing to its benign behavior, slow growth and clear delineation, as well as its low tendency to recur (0.2%), the treatment of choice is conservative surgical enucleation and simple curettage.[16] Similar approach was practiced in our case and the patient had no recurrence, and regular follow-up was done after local excision.

Conclusion

Thus, based on currently available evidence and the findings in the present case we emphasize that extraosseous/peripheral variant of AOT is very rare. Here, we have discussed the clinical, radiographic and histopathologic features of a case of peripherally occurring AOT, which involved left anterior maxilla of a young male.

It was initially diagnosed as an odontogenic cyst and was conservatively surgically enucleated. The final diagnosis of an AOT was arrived by histologic examination of the excised tissue specimen. Recurrence of AOT is exceptionally rare. Hence, the prognosis is excellent.

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