Case Report - Pathology

Maxillary adenomatoid odontogenic tumor associated with a premolar

Access this article online
Website: www.amsjournal.com
DOI: 10.4103/2231-0746.161118
Quick Response Code:

Vimal Kalia, Geeta Kalra, Nitin Kaushal[1], Vikas Sharma[2], Mayank Vermani

Departments of Oral, Maxillofacial Surgery and [1]Pathology, BRS Dental College and Hospital, [2]Consultant, Dantantra, H No 731, Sector 2, Panchkula, Haryana, India

Address for correspondence:
Dr. Vimal Kalia, Department of Oral and Maxillofacial Surgery, BRS Dental College and Hospital, Dantantra, House No: 731, Sector 2, Panchkula, Haryana, India.
E-mail: vimal@kalia.in

ABSTRACT

The adenomatoid odontogenic tumor (AOT) represents 3–7% of all odontogenic tumors, and over 750 cases have been reported in the literature. This lesion was formerly considered to be a variant of the ameloblastoma and was designated as adenoameloblastoma. These lesions may infrequently produce dentinoid material and rarely enamel matrix. Consequently, the WHO (2005) classification of odontogenic lesions considered this process to represent a mixed odontogenic neoplasm. We present a case of a 12-year-old female patient with an AOT of diameter 5 cm × 5 cm located in the anterolateral region of the maxilla in association with an impacted premolar tooth. The rarity of AOT, association of this lesion with regards to maxillary premolar, the exaggerated size at presentation, the eruption of the displaced canine postoperatively and uneventful healing of the bony defect without adjunctive therapy makes this case unique.

Keywords: Adenomatoid odontogenic tumor, ameloblastoma, impacted teeth, odontogenic tumor

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a rather uncommon tumor of odontogenic origin.[1] It is a slow-growing, benign, epithelial tumor with clinical presentation of absence or impaction of teeth, absence of pain, expansion of bone and deformity of the face as the lesion develops and grows.

The term "AOT" was introduced by Philpsen and Birn and later adopted by the WHO (2005) in their "Histological typing of odontogenic tumors, jaw cysts and allied lesion."[2] The etiopathogenesis of AOT is still unclear and controversial. AOT is considered as a developmental outgrowth or a hamartoma while others consider it as a major neoplastic growth of odontogenic epithelium.[3,4]

Adenomatoid odontogenic tumor was classified by the WHO as a mixed odontogenic neoplasm because it sometimes contains dental hard tissue formed by interactions between the dental epithelium and mesenchyme. Presently AOT is classified as an epithelial tumor without odontogenic ectomesenchyme and falls under the category of tumor with "intermediate frequency" that is, between 1% and 10%.[2,5] Histopathologically, AOT is a pure epithelial odontogenic tumor without induction and has an overall distinctive histomorphology which exhibits a remarkable consistency making its diagnosis easy and nonambiguous.[4,6]

We report a 12-year-old female patient with an AOT of size 5 cm × 5 cm located in the anterolateral region of the maxilla in association with an impacted premolar tooth. An impacted and displaced canine was found outside the lesion which regained its path of eruption after enucleation of the AOT in the follow-up period. The rarity of AOT, detailed radiographic analysis, association of this lesion with regards to its maxillary premolar, the exaggerated size at presentation, the eruption of the displaced canine in postoperative period and uneventful healing of the bony defect without adjunctive therapy makes this case unique.

CASE REPORT

A 12-year-old female reported to the maxillofacial surgery clinic with the complaint of a painless mass in the left buccal area of

the maxilla. Furthermore, the patient complained of absence of the maxillary canine and first premolar tooth on the same side since 2 years.

On extraoral examination, there was a swelling on the left side of the face causing obvious asymmetry with obliteration of nasolabial sulcus. Intraorally, there was an expansion in all dimensions measuring about 5 cm × 5 cm in anteroposterior and superior inferior direction. On palpation, the swelling was hard, nonlobulated; nontender, not fixed to the overlying skin and local temperature was not raised. The growth had expanded the palatal, alveolar and buccal aspect to such an extent that the overlying mucosa had indentation marks from the lower teeth causing discomfort to the patient during speech and mastication [Figure 1].

The orthopantomography (OPG) revealed [Figure 2] a radiolucent lesion with irregular borders located between the apices of the left maxillary lateral incisor and the left maxillary second premolar. The canine was displaced toward the apices of the incisors, and the first premolar tooth was present within the lesion. The coronal computed tomography (CT) of the maxilla revealed a well-circumscribed radiolucent mass with calcific bodies which were multiple and interspersed throughout the lesion with prominent sclerotic margins. Maxillary left canine was impacted without being involved in the lesion. The axial CT confirmed that the left maxillary sinus and the nasal cavity were not encroached. Maxillary left first premolar was present within the lesion [Figure 3].

Under premedication (i.e. injection atropine, 0.6 mg IM, 60 min before surgery and injection diazepam 5–10 mg IM) and local anesthesia, an adequate window was created and the tumor mass was enucleated along with the enclosed 24 tooth [Figures 4 and 5]. The mass was partially solid with partial cystic degeneration, and a gritty sensation could be elicited on examination. The remaining cavity was found to be clean without any tissue tags hence chemical or mechanical curettage was not done. The wound was sutured with 3–0 silk with an open packing. The packs were changed after weekly intervals for 6 weeks and the patient was instructed to keep the area clean.

The histopathological picture [Figure 6] of the biopsy sample revealed epithelium and underlying connective tissue stoma. The tumor cells were seen arranged in various patterns firstly, duct-like structures of

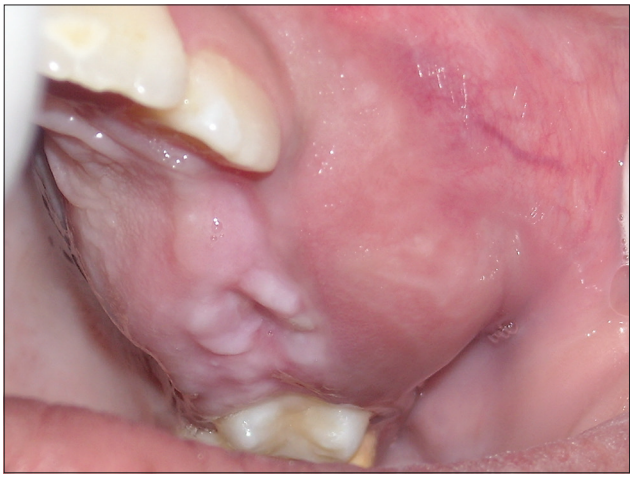

Figure 1: Preoperative intraoral picture

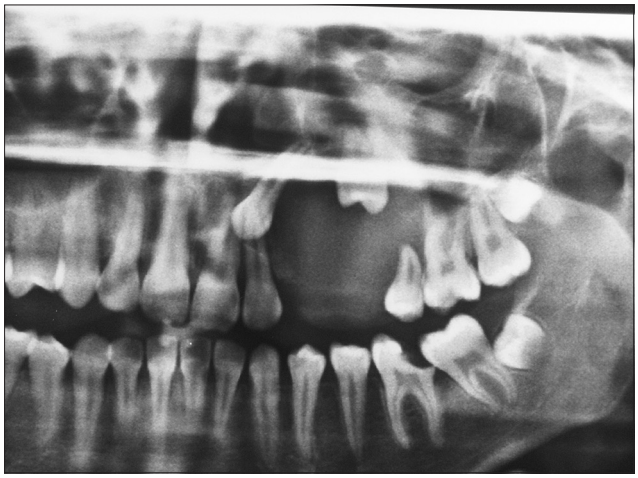

Figure 2: Preoperative orthopantomograph

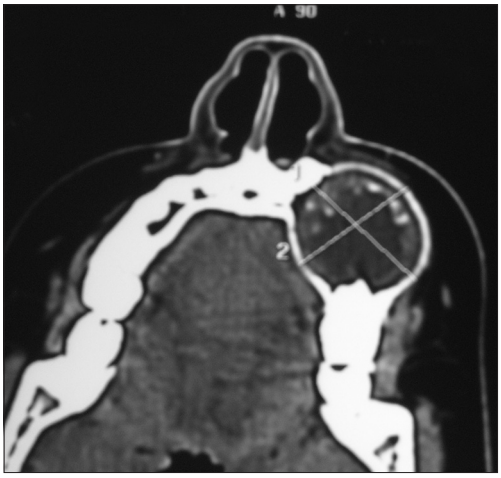

Figure 3: Preoperative computed tomography scans (axial view)

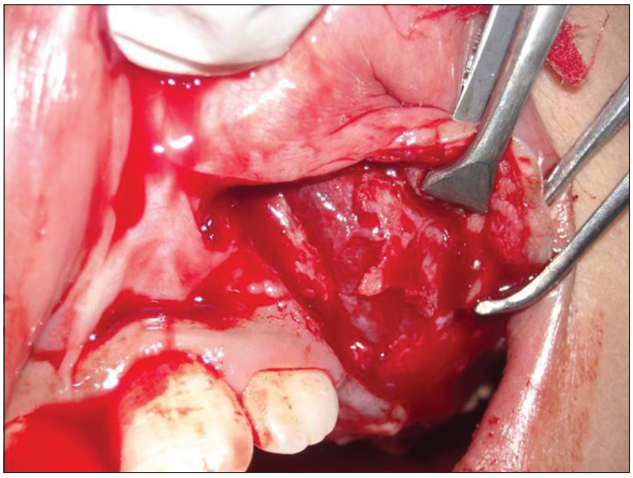

Figure 4: Intra-operative picture

varying sizes which were lined by cuboidal cells with a lumen lined by an eosinophilic rim of varying thickness. Secondly, rosettes like structures lined by tall columnar cells with hyperchromatic polarized nuclei were seen [Figure 7]. The connective tissue also showed the presence of multiple calcified structures along with amorphous eosinophilic material at a few places. The histopathologic diagnosis confirmed the lesion to be a follicular AOT.

During the 1-year follow-up period both clinically and radiographically, we observed a gradual reduction in the size of the deformity and adequate healing of the defect. The displaced maxillary left canine also moved on the eruption path and axis [Figure 8].

DISCUSSION

Adenomatoid odontogenic tumor is a rare, benign, nonaggressive epithelial tumor which exhibits slow but progressive growth, derived from the epithelial component of the tooth forming tissues, but none of the associated teeth were described as morphologically defective.Thus, the disturbance must occur after odontogenesis is complete.[7,8] Stafne originally suggested that the cell of origin was that of the epithelium entrapped in the line of embryonal fusion, but the current belief is that it originates from the odontogenic epithelium of the dental lamina complex or its remnants.[9]

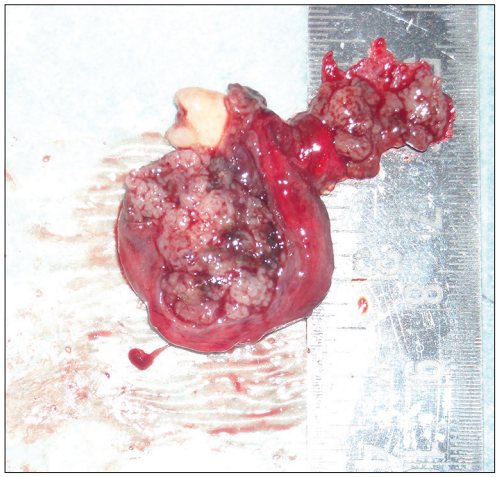

Figure 5: Enucleated mass

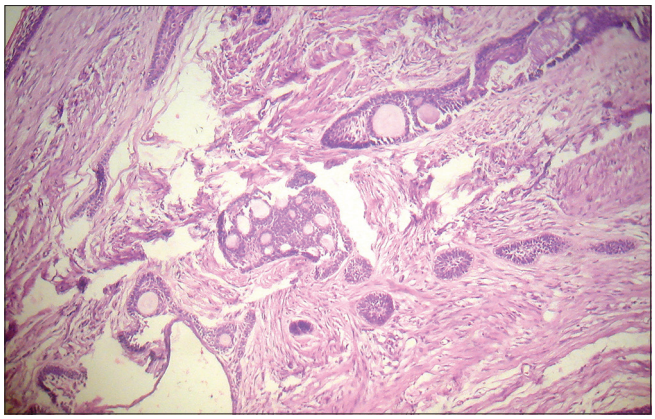

Figure 7: Pictomicrograph (H and E, × 45)

About 53% of AOT's occur in the anterior maxilla, and 9% occur in the maxillary premolar region. 2% AOT's occur in the molar region.[10] Unerupted permanent tooth/teeth are associated with the lesion in 1/3rd of the cases[2,11] and the follicular variant of the AOT, which is far more common variant (75%) is associated with the crown of an impacted tooth, most commonly, the maxillary canine.[3] Being a painless growth, the tumor acquires larger dimensions, and the unusual size may be 1.5–3 cm at presentation. There has been a similar case report in the literature by Olgaç *et al*. of an AOT with a diameter 3.5 cm × 4 cm located in the anterior region of the maxilla in which an impacted premolar was detected and a displaced canine was found outside the lesion. In our case, the lesion was similar but larger in size.[12]

Radiographically, AOT has two variants, that is, follicular and extrafollicular type. The follicular variety, as in the present case shows a well-defined, unilocular (round or ovoid) radiolucency associated with either the crown of the tooth, the root or the whole tooth,[10] thus mimicking a dentigerous cyst or in the presence of scattered radiopacities mimicking a calcifying epithelial odontogenic cyst or tumor. In a few cases, more than one unerupted tooth was associated with the tumor.[2,13] In the present case, premolar was enclosed in the lesion whereas canine was displaced, impacted and not enclosed in the AOT. In the presence of the radiopaque spots, the differential diagnosis should also include fibrous dysplasia, pindborg tumor, ameloblastic odontofibroma and ghost cell odontogenic cyst.[14] Hence, radiographic differentiation of such odontogenic lesions becomes a difficult task. Diagnosis can be established by careful analysis

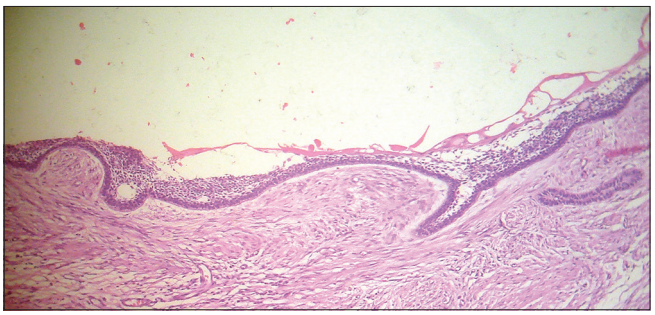

Figure 6: Pictomicrograph (H and E, ×10)

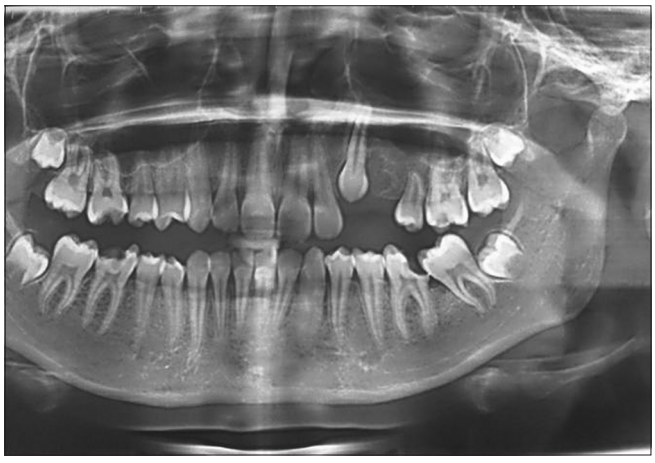

Figure 8: Follow-up orthopantomography showing corrected eruption axis of canine

of the site of the lesion, its borders and effect of the lesion on adjacent structures, presence and extension of cortices, erosion of cortices of anatomic structures, root divergence, root resorption and calcific bodies in the various radiographic presentations of the AOT.

An OPG does give a comprehensive view of the lesion, but a multi-slice CT scan provides the extension of the lesion in all directions besides showing dental involvement, as in this case it revealed extracapsular presence of canine tooth which could hence be saved and allowed to erupt. It also showed noninvolvement of the maxillary sinus and aided in treatment planning. Quality and quantity of the scattered radiopaque foci can also be well appreciated on the CT scan. CT scan provides proper extensions of the AOT, reveals proximity to the vital structures which aid in the preoperative evaluation, provisional diagnosis and the planning and execution of the surgical procedure.

Histologically, all variants of AOT show a consistent and identical picture. They are characterized by odontogenic epithelium with duct-like structures with varying degrees of inductive changes in the connective tissue as described by WHO.[4,15] Tatemoto *et al*. suggested expression of keratin and vimentin in the tumor cells at the periphery of the ductal, tubular and whorled structures.[16] Furthermore, Leon *et al*. in their study found 27 cases positive for vimentin.[17] Dystrophic calcification, osteodentin and cementum like substance has been reported by some authors whereas the occurrence of enamel matrix is extremely rare.[4,18] In intrafollicular variety, a definitive diagnosis is accomplished by surgical exploration as the encapsulation makes enucleation easy. The residual clean cavity and grossing of the enucleated mass along with the tooth and gritty areas due to calcifications help predict it as an AOT.

In the treatment of AOT, most authors now believe that complete enucleation with long-term observation is the preferable treatment. Since all variants of AOT show identical, benign biological behavior and since most of the cases are well encapsulated, conservative surgical enucleation has proved to be the treatment of choice with rare recurrence.[4,16,17] Though AOT is not known to recur, but Toida *et al*. reported two cases, with recurrences, and one of them had intracranial extension.[11] Philipsen and Reichart reported recurrence in only three cases out of 750 studied.[4] Long-term follow-up is thus necessary to observe the complete filling in of the residual bony defect, fate of the involved tooth/teeth and recurrences if any. Guided tissue regeneration in conjunction with bone grafting can be used as an aid for rapid filling of large defects surrounding teeth created by odontogenic tumors.[6] In our opinion, unerupted tooth can be given a fair chance of eruption. Bone graft should not be placed so that intact periosteum with adequate closure shall bring optimum bone healing as is evident in this report.

In our case, the lesion being larger and follow-up being longer, we could observe the corrected deformity, the canine tooth had regained its path of eruption and the bony cavity had healed optimally in the absence of any adjunctive therapy. The patient was referred for orthodontic intervention for guided eruption of the canine and later implant supported rehabilitation of premolar is planned.

CONCLUSION

The rarity of AOT can be the reason that it is not known in its entirety. Pathologic entities have a varied presentation with regards to size, location, sex predilection, clinical and radiographic presentation making diagnosis and treatment planning a challenge.

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Cite this article as: Kalia V, Kalra G, Kaushal N, Sharma V, Vermani M. Maxillary adenomatoid odontogenic tumor associated with a premolar. Ann Maxillofac Surg 2015;5:119-22.

Source of Support: Nil, **Conflict of Interest**: None declared.