

# Adenomatoid Odontogenic Tumor of Follicular Variant Affecting the Anterior Maxilla: A Case Report

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## Abstract

Adenomatoid odontogenic tumor (AOT) is uncommon lesion of odontogenic origin which is benign and non-invasive. Young female patients are commonly affected. Lesion is slow growing, asymptomatic, and mainly involved maxillary anterior region. In this case report, this lesion is of follicular variant and is in association with maxillary impacted 13. Enucleation and curettage are the treatment of choice. There is 0.1% recurrence rate observed in this lesion. However, in this case, we did 3 years follow-up of the patient, but reoccurrence was not observed. The early intervention and diagnosis can prevent patient's esthetics and stops further destruction of adjacent structure. Any swelling of longer duration associated with deciduous teeth present in maxillary anterior region should be suspected as odontogenic lesion.

**Key words:** Follicular AOT, Adenomatoid odontogenic cyst, Hamartomas

## INTRODUCTION

Steensland, in 1905, considered adenomatoid odontogenic tumor (AOT) lesion as a variation of ameloblastoma.<sup>[1]</sup> AOT accounts for 3–7% of all odontogenic tumors.<sup>[2]</sup> In 1948, Stafne considered it as a separate entity.<sup>[2]</sup> In 1971, the World health organization (WHO) accepted the term AOT.<sup>[3]</sup> Philipsen *et al.* classified AOT into types follicular, extrafollicular, and peripheral.<sup>[4]</sup> This lesion is slow growing, non-invasive, and asymptomatic. This lesion is common in the 2<sup>nd</sup> and 3<sup>rd</sup> decades of life, which accounts for 88% cases.<sup>[5]</sup> Female-to-male ratio is 2:1 and affects upper anteriors.<sup>[6]</sup> Lesion has very distinct capsule surrounding it and structures resembling ducts (adenomatoid) within epithelial lesion.<sup>[7]</sup> It rarely reoccurs even if it is treated with a conservative curettage.

## CASE REPORT

A 11-year-old female patient reported to our department with chief complaint of swelling in the right maxillary front region since 1 year. Swelling was painless, initially small in size gradually grew to present size. There was no relevant medical history and history of trauma.

On extraoral examination [Figures 1-3], facial asymmetry was present. On the right side of maxilla, there was a solitary, localized, and spherical-shaped swelling approximately 4 × 4 cm in size present with center 1 cm posterior to alae of the nose obliterating nasolabial fold. Overlying skin was normal, movable, and not fixed with the swelling suggestive of intraoral swelling without involving skin. The swelling was non-tender and bony hard in consistency. Anteroposteriorly, the swelling was extending from alae of nose to perpendicular line drawn from outer canthus of eye. Superoinferiorly, the swelling was extending from 1 cm inferior to infraorbital margin until commissure tragus line.

On intraoral examination [Figure 4], the swelling was present in the right maxillary buccal region extending

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**Month of Submission :** 04-2022  
**Month of Peer Review :** 05-2022  
**Month of Acceptance :** 05-2022  
**Month of Publishing :** 06-2022

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Figure 1: Shows facial asymmetry on right side of maxilla



Figure 2: Shows extraoral swelling present on the right side of maxilla, there is a solitary, localized, and spherical-shaped swelling approximately 4 × 4 cm in size present with center 1 cm posterior to alae of the nose obliterating nasolabial fold



Figure 3: Shows extraoral swelling present on the right side of maxilla, there is a solitary, localized, and spherical-shaped swelling approximately 4 × 4 cm in size present with center 1 cm posterior to alae of the nose obliterating nasolabial fold

from mesial margin of 11 until mesial margin of 16. Superoinferiorly, it was from gingival margin to the depth of the vestibule causing vestibular obliteration. Overlying mucosa was normal in color. Swelling was hard in consistency suggesting expansion of buccal cortex.



Figure 4: Intraoral swelling present on right maxillary buccal region extending from mesial margin of 11 until mesial margin of 16

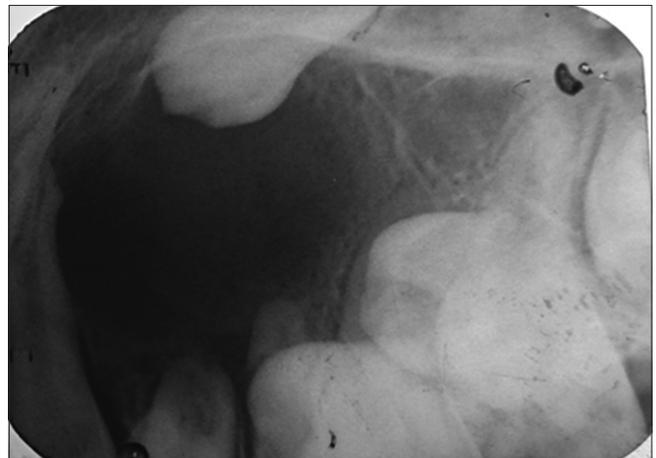


Figure 5: IOPA shows a well- defined radiolucency in periapical region extending from mesial border of 12 without involving apex of 11 until mesial border of 16



Figure 6: Superoinferiorly, radiolucency was extending from crest of alveolar bone of 12, 53, 54, and 55 region to lifting floor of the maxillary sinus. 13 was impacted and superiorly displaced. Root apex of 12 was mesially tilted causing space between crown of 11 and 12. The right central incisor was slightly extruded. Root resorption was seen in 53 and grossly carious 54

Aspiration was found negative. Electric vitality test of 12 demonstrated negative response. Provisional diagnosis of Adenomatoid odontogenic tumor with differential diagnosis of dentigerous cyst was considered.

IOPA and OPG [Figures 5 and 6] showed a well-defined radiolucency in periapical region extending from mesial border of 12 without involving apex of 11 until mesial border of 16 anteroposteriorly. Superoinferiorly, radiolucency was extending from crest of alveolar bone of 12, 53, 54, and 55 region to lifting floor of the maxillary sinus. Thirteen was impacted and superiorly displaced. Root apex of 12 was mesially tilted causing space between crown of 11 and 12. The right central incisor was slightly extruded. Root resorption was seen in 53 and grossly carious 54. Roots of developing premolars were pushed posteriorly causing anterior inclination of crowns.

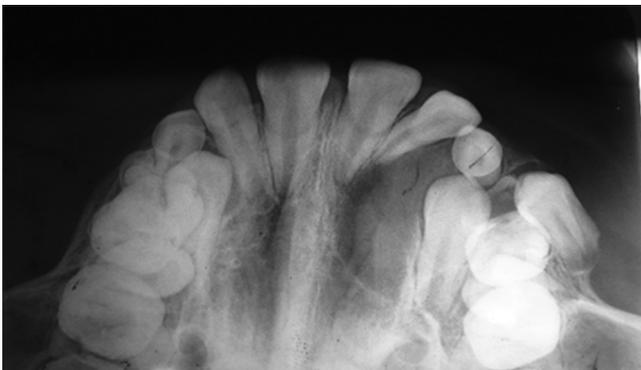
Occlusal view [Figure 7] showed a well-defined corticated radiolucency causing buccopalatal expansion without displacing nasal septum. Cone-beam computed tomography

[Figures 8 and 9] showed a well-defined corticated expansile lesion approximately  $4.5 \times 3.5$  cm lifting the anteroinferior floor of maxillary sinus without perforating it. Thirteen was pushed superoanteriorly and completely enveloped by the lesion. Lesion was expanded until lateral wall of the nose without its displacement or perforation.

Lesion was surgically enucleated [Figures 10 and 11] with extraction of 13, 53, and 54. Based on clinical, radiological, and histopathological findings, a diagnosis of Adenomatoid odontogenic tumor was made. The patient visited to the department after 12 months [Figure 12]. The patient is still under follow-up.

## DISCUSSION

AOT has been given variety of terminologies such as adenoameloblastoma, ameloblastic adenomatoid tumor, odontogenic adenomatoid tumor, and pseudoadenoma adamantinum.<sup>[8]</sup> AOT occurs 2/3<sup>rd</sup> in young females, maxilla,



**Figure 7:** Occlusal view showed a well defined corticated radiolucency causing buccopalatal expansion without displacing nasal septum



**Figure 9:** CBCT showed a lesion was expanded until lateral wall of the nose without its displacement or perforation



**Figure 8:** CBCT showed a well defined corticated expansile lesion approximately  $4.5 \times 3.5$  cm lifting the anteroinferior floor of maxillary sinus without perforating it



**Figure 10:** Extracted 13, 53, 54



**Figure 11: Lesion was surgically enucleated and 13, 53, 54 were extracted**



**Figure 12: Post op follow up after 1 year**

and its association with unerupted teeth especially canine, so it is also known as “two-thirds tumor.”<sup>9]</sup> About 73% cases of AOT are follicular type, in which there is central lesion in association of impacted tooth as seen in this case; 24% of cases are extra follicular type, in which there is no connection of central lesion with the tooth; and 3% of cases are of peripheral type that is present in gingival tissue of tooth bearing areas.<sup>14]</sup> Odontogenic sources such as enamel organ, reduced enamel epithelium, dental lamina, and their remnants are thought to be the cause of AOT. Features of AOT include asymptomatic nature, cortical expansion which is present often and lesion involving the tooth is commonly impacted with slight displacement of adjacent tooth. All these features mentioned were seen in this case.

AOT shows resemblance with many odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, ameloblastomas, odontogenic keratocysts, and calcifying epithelial odontogenic tumor. It can be distinguished from dentigerous cyst as radiolucency circumscribing the tooth shows apical displacement from CEJ, while in dentigerous cyst, it never crosses CEJ.<sup>10]</sup> Histopathologically, AOT shows spindle-shaped or polygonal cells which form sheets and whorled masses in scarce connective tissue stroma. Duct such as structures is lined by single row of columnar epithelial cells and nuclei are polarized away from central lumen. Dystrophic calcification is mostly seen in AOT.<sup>11,12]</sup>

## CONCLUSION

Impacted permanent teeth or retained deciduous teeth for longer duration when associated with swelling should always be suspected for odontogenic lesion.

## ACKNOWLEDGMENT

General support by assisting staff.

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**How to cite this article:** Bathula H, Yeso A, Garg A, Shreeja R, Suri N, Singh SK. Adenomatoid Odontogenic Tumor of Follicular Variant Affecting the Anterior Maxilla: A Case Report. *Int J Sci Stud* 2022;10(3):1-4.

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