

Case Series: Adenomatoid Odontogenic Tumor

Dr. B.C. Sikkerimath¹, Dr. Aditya Anshu², Dr. Anu Jose³

¹Professor and HOD, Dept of Oral and Maxillofacial Surgery, PMNM Dental College and Hospital, Bagalkot, India

^{2,3}Resident, Dept of Oral and Maxillofacial Surgery, PMNM Dental College and Hospital, Bagalkot, India

Abstract: Adenomatoid odontogenic tumors are rare, benign, non invasive lesions. AOT commonly occurs in anterior maxilla, common in females of younger age and commonly associated with impacted canine. It shows clinical and radiographical similarities to other odontogenic cysts like dentigerous cysts. They can be definitely diagnosed with the help of histopathological examination and it is important to confirm the diagnosis to prevent mutilating surgeries. AOT can be treated with simple enucleation and curettage with low rate of recurrence (0.2%). Here we present six cases of Adenomatoid odontogenic tumor treated with surgical enucleation and curettage.

Keywords: Enucleation, Adenomatoid odontogenic tumor, two-third tumor, follicular type

1. Introduction

Adenomatoid odontogenic tumor (AOT) is a rare, benign odontogenic lesion which is usually slow growing, painless and non invasive. It was first recognized by Stafne in 1948 and the name AOT was proposed by Philipsen and Birn in the year 1969[4],[7]. It represents 3 to 7% of all odontogenic tumors [3]. The World health organization (WHO) in 1971 defined AOT as a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue[1]. AOT is also known as 'two-third tumor' because about two-third cases occur in the maxilla, about two-third cases arise in young females, two-third cases are associated with an unerupted tooth, and in two-third cases affected teeth are canines.[5],[6]. Clinically and radiographically, often it is misdiagnosed as odontogenic cysts like dentigerous cyst [2]. However radiographically, fine calcifications which give snowflake appearance in AOT helps in differentiating them. Here we present 6 cases of AOT which are treated by surgical enucleation and curettage.

Case 1

A 19 year old female patient presented with chief complaint of swelling in upper left front midface region since 4 months. Initially the patient was apparently normal when she noticed the swelling which was initially of peanut size, which gradually increased in size and reached the present state.

On extraoral examination, a solitary swelling is noted on the left midface region measuring around 4 cm x 5 cm in maximum dimension extending from the left ala of nose till the line joining lateral canthus of left eye anteroposteriorly. On palpation it is firm to hard in consistency and non-tender.



Figure 1: Preoperative photograph

On intraoral examination, diffuse swelling is seen extending from left maxillary permanent lateral incisor to left maxillary permanent first molar region. Also vestibular obliteration noted. Mucosa over the swelling appears normal. On palpation, inspector findings are confirmed. It is non tender and hard in consistency.

3 D CT face reveals expansile lesion on left side of face extending from infraorbital rim to roots of left maxillary permanent incisor to left maxillary permanent first molar region with impacted left maxillary permanent canine.



Figure 2: Preoperative 3D CT Face

It is treated with surgical enucleation. The cystic lining is removed along with the impacted tooth. Irrigated with gentamycin and sutured with 3-0 vicryl.



Figure 3: Intraoperative photographs

Case 2

A 19 year old female presented with chief complaint of swelling in left mid-face region since 5 months. The swelling was initially small in size, which gradually increased and has reached the present state.

On extraoral examination, diffuse swelling is seen in left midface region. Mild facial asymmetry noted.

On intraoral examination, swelling is noted that extends from permanent maxillary left central incisor to permanent

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maxillary left first premolar region. Vestibular obliteration noted. Mucosa appears normal over the swelling.

Orthopantomogram (OPG) reveals well defined expansile cystic lesion involving left maxilla and one unerupted tooth.

3 D CT face reveals expansile lesion on left side of face extending from infraorbital rim to roots of left maxillary permanent incisor to left maxillary permanent first molar region with impacted left maxillary permanent canine.

It was provisionally diagnosed as a tumor of odontogenic origin and was treated by surgical enucleation under general anesthesia.



Figure 4: Intra-operative photographs

Case 3

A 27 year old female patient presented with chief complaint of pain and swelling in upper left half of face since 7 months. Patient gives history of trauma by sugar cane after which the swelling started appearing. It was initially of small peanut size, which gradually increased and reached to present state. There is no associated pain or complaints.

On extraoral examination, mild swelling noted in left side of face. On palpation, the swelling is tender and firm to hard in consistency.

On intraoral examination, diffuse swelling is seen extending from left maxillary permanent canine to left maxillary permanent first molar region. vestibular obliteration noted and is tender on palpation.



Figure 5: Preoperative photographs

OPG reviews unicystic radiolucency with respect to upper anterior maxilla on the left side.

It was provisionally diagnosed as AOT and was treated by surgical enucleation.



Figure 6: Intra-operative photographs

Case 4

A 12 year old female patient presented with chief complaint of swelling in her right midface region since 3 months. Swelling was small in size initially which gradually increased in size and reached to present state. There is no associated pain, difficulty in chewing or mouth opening, pus discharge or fever.

On extraoral examination, diffuse swelling is seen in right midface. On palpation, the swelling is firm to hard in consistency, non fluctuant and non tender.

On intraoral examination, vestibular obliteration is seen in relation to maxillary right central incisor to maxillary right second premolar region. On palpation, swelling is firm to hard in consistency and is non tender. Preshedding mobility noted with respect to right deciduous canine.

OPG reveals well defined radiolucent area with well corticated border.

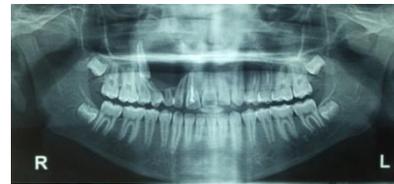


Figure 7: Preoperative OPG

3 D CT face reveals expansile lesion on right side of face extending from infraorbital rim to roots of right maxillary permanent central incisor to right maxillary permanent first molar region with impacted left maxillary permanent canine.

The tumor mass was removed in toto along with the cystic lining and impacted canine. Irrigated with gentamycin and closure done with 3-0 vicryl.



Figure 8: Intra-operative photographs

Case 5

A 26 year old female patient presented with chief complaint of swelling in her right lower third of face since 1 month. Swelling was initially small which gradually increased in size and reached to present state.

On extraoral examination, diffuse swelling is seen in right lower third of face. On palpation, the swelling is firm to hard in consistency, non fluctuant and non tender.

On intraoral examination, vestibular obliteration is seen in relation to mandibular right lateral incisor to mandibular right second premolar region. On palpation, swelling is firm to hard in consistency and is non tender.

OPG reviews well defined radiolucent area with well corticated border.

It was provisionally diagnosed as AOT and was treated by surgical enucleation under general anesthesia.



Figure 9: Intra-operative photographs

Case 6

A 15 year old female patient presented with chief complaint of swelling in upper left front midface region since 6 months. The swelling was initially of 1 x 1 cm size, which gradually increased and reached the present state.

On extraoral examination, a swelling is noted on the left midface region. On palpation, it is firm to hard in consistency and non-tender.

On intraoral examination, swelling is present extending from midline to distal aspect of maxillary left permanent first molar region. Vestibular obliteration noted. Also preshedding mobility present with respect to deciduous left lateral incisor and deciduous left canine. On palpation it is firm to hard in consistency and is non-tender.



Figure 10: Preoperative photographs

OPG reviews unicystic radiolucency with respect to upper anterior maxilla on the left side.

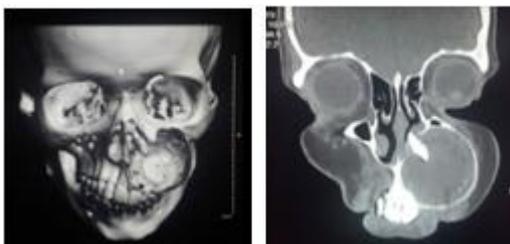


Figure 11: Preoperative 3D CT Face

3 D CT face reveals expansile lesion on left side of face extending from infraorbital rim to roots of left maxillary permanent incisor to left maxillary permanent first molar region with impacted left maxillary permanent canine.



Figure 12: Intraoperative photographs

It is treated with surgical enucleation along with cystic lining and impacted tooth. Irrigated with gentamycin and sutured with 3-0 vicryl.

Histopathological Examination

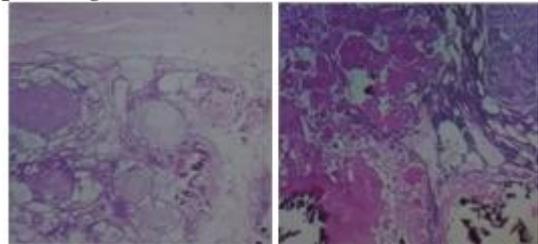


Figure 13: Histopathology report

The H & E stained and studied section in all six cases shows well capsulated odontogenic cystic lesion. The lesional cells are oval to spindle shaped darkly staining cells suggestive of odontogenic epithelial cells having vesiculated nuclei, arranged in various patterns like whorls, solid nodule, strand like, duct like areas and plexiform pattern in the periphery. Areas of acellular eosinophilic material and abundant round to irregular areas of calcifications are also evident distributed throughout the lesion. Hemorrhagic areas also seen.

2. Discussion

In 1971, World Health Organization classification of odontogenic tumors mentioned AOT as a mixed odontogenic neoplasm, that is as an epithelial tumor with an inductive effect on the odontogenic mesenchyme [10].

It is a rare, benign lesion which commonly occurs in anterior maxilla and is routinely detected in radiographs. It usually manifest as an asymptomatic swelling which is non invasive. It commonly occurs in tooth bearing areas of jaw and is usually associated with an impacted teeth [8].

AOT is detected during a routine radiographic examination or when radiographs are made to examine an unerupted tooth [3, 10]. Three variants of AOT are described a) Follicular type has an intra-bony lesion associated mostly with an impacted tooth and is usually misdiagnosed as a dentigerous cyst or follicular cyst. b) Extrafollicular type has an intra-bony lesion but is not associated with a tooth. Usually present as a well defined unilocular radiolucent area which is above or superimposed on the roots of the erupted teeth. c) Peripheral type usually presents as a gingival swelling and often appears as a small, sessile mass on the maxillary gingiva [11].

In this series, all the cases were of follicular type (associated with an impacted tooth). There were no peripheral AOTs. The maxilla was most commonly involved with the most common impacted tooth being the canine. In the mandible, the canine and premolars were equally affected. The above findings are consistent with the literature[12].

Clinically and radiographically it shows similarity to other odontogenic cysts which makes it difficult to diagnose. However Dare et al. found that the intraoral periapical radiograph is the best radiograph to show radio-opacities in

AOT as discrete foci having a flocculent pattern within radiolucency even with minimal calcified deposits giving it a snowflake appearance [8].

Radiographically, the lesions reported in this case series appeared as well-demarcated unilocular radiolucencies associated with an unerupted tooth. No radiopacities were noted within the radiolucency. Philipsen and Reichart reported that approximately two-thirds of the intrabony cases have scattered radiopacities within the radiolucency[12].

Surgical management that is enucleation along with the removal of associated impacted tooth and simple curettage is the treatment of choice. The literature reports recurrence rate as low as 0.2% after enucleation and curettage [8],[9]. In the cases described here, follow up is done for upto 6 months with no any evidence of recurrence.

3. Conclusion

AOT shows clinical and radiographic similarities to many other odontogenic cysts. Because of its benign and non invasive nature, AOT can be conservatively treated with enucleation and curettage with less chances of recurrence. However histopathologically, other odontogenic cysts should be ruled out to avoid mutilating surgeries.

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Author Profile



Dr. Aditya Anshu, PG Resident, Department of Oral and Maxillofacial Surgery, PMNM Dental College and Hospital, Bagalkot, Karnataka



Dr. Anu Jose, PG Resident, Department of Oral and Maxillofacial Surgery, PMNM Dental College and Hospital, Bagalkot, Karnataka