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IP Journal of Otorhinolaryngology and Allied Science

Journal homepage: <https://www.joas.co.in/>

## Case Report

# Adenomatoid odontogenic tumour of maxillary canine presenting as a dentigerous cyst: A case report

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## ARTICLE INFO

### Article history:

Received 11-09-2023

Accepted 20-10-2023

Available online 27-10-2023

### Keywords:

Adenomatoid odontogenic tumor

Dentigerous cyst

Ameloblastoma

Caldwell luc operation

Enucleation

## ABSTRACT

Our article reports the case of a adenomatoid odontogenic tumor (AOT) associated with an upper permanent central incisor, previously diagnosed as dentigerous cyst. The condition is a slow-progressing and nonmalignant epithelial tumor. Specifically, it mentions that it is an Adenomatoid Odontogenic Tumor (AOT). The clinical features of AOT are outlined, including its association with impacted teeth or, in the absence of impacted teeth, it presents as painless, bony expansion and facial asymmetry due to the growth of the lesion. A 16-year-old female patient presented to the Department of ENT with a chief complaint of swelling on the upper left side of the face that had been present for 2 months. The patient underwent a surgical procedure, specifically a Caldwell luc operation and left medial maxillectomy with Denker's technique. The pathology reports confirmed the diagnosis of a benign odontogenic tumor favoring Adenomatoid Odontogenic Tumor. The article emphasizes the importance of early diagnosis and treatment in children to achieve the best possible outcome in managing such conditions.

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## 1. Introduction

AOT is an uncommon tumor with odontogenic origins. It was first described by Steensland in 1905. Subsequently, in 1907, Dreibradt introduced the term "pseudo ameloblastoma" to describe this condition. AOT is often associated with impacted teeth. In other words, it may develop around unerupted teeth, leading to its discovery during dental examinations or when investigating the cause of delayed tooth eruption. AOT is typically painless, which means it doesn't cause significant discomfort or pain for the patient.<sup>1</sup> This is one reason why it can go unnoticed until it leads to other clinical manifestations. OT can result in bony expansion of the affected area. This means that as the tumor grows, it can cause the surrounding bone to expand or become thicker, which can be observed through imaging studies like X-rays. The growth of the AOT lesion can lead

to facial asymmetry. This is particularly noticeable when the tumor develops in the maxilla (upper jaw) or mandible (lower jaw), causing changes in the shape or appearance of the face.<sup>2</sup> This lesion is known by many names, including adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantium, or teratomatous odontoma. AOT is classified into three main variants based on its location and presentation: Follicular Variant, Extrafollicular Variant, Peripheral Variant.<sup>3</sup>

The dentigerous cyst is one of the most common types of odontogenic cysts, accounting for approximately 33% of all odontogenic cysts. It is often associated with an impacted tooth, and it typically develops after the complete formation of the tooth's crown. dentigerous cysts typically form around the crown of an impacted tooth, leading to a well-defined radiolucent lesion in the jawbone. These cysts are usually associated with unerupted or impacted teeth, and they can cause various dental complications if left untreated.

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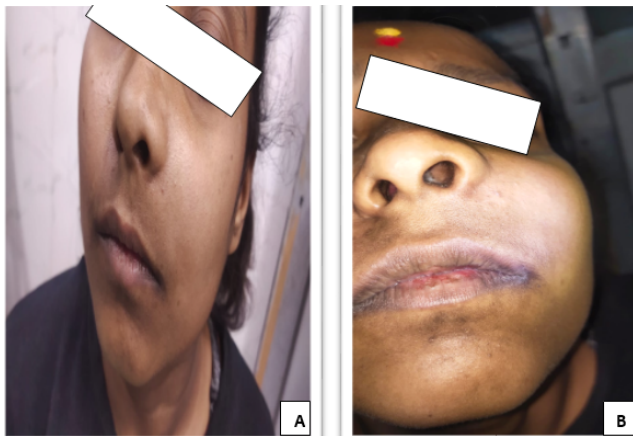
The mandibular third molar (wisdom tooth) is indeed the most commonly affected tooth, followed by the maxillary canine (cuspid) and mandibular premolars. These teeth are more prone to becoming impacted or delayed in their eruption, which increases the likelihood of a dentigerous cyst forming around them. Dentigerous cysts do tend to progress slowly, and they are often asymptomatic in the early stages. They are frequently discovered incidentally during routine radiographic examinations such as dental X-rays.<sup>4</sup>

## 2. Case Report

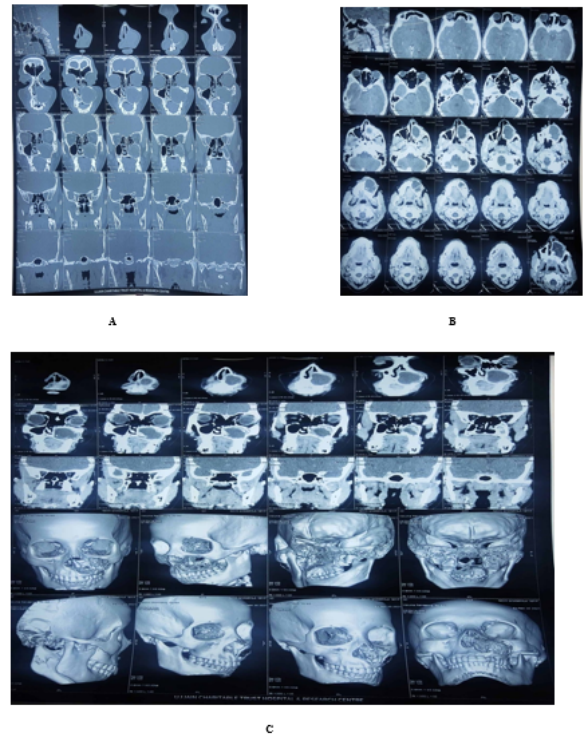
A 16-year-old female patient reported to the Department of ENT, with the chief complaint of swelling in the upper left side of the face for 2 months. Initially, the swelling was pea sized and gradually increased to its current size. The extraoral examination revealed oval shaped facial swelling measuring approximately 5 cm × 5 cm. On palpation, the swelling was well defined, firm to hard consistency, non-tender with no local rise in temperature, non-pulsatile and non-fluctuant. A little bulge on the floor of left nasal cavity was also seen.

### 2.1. Investigations

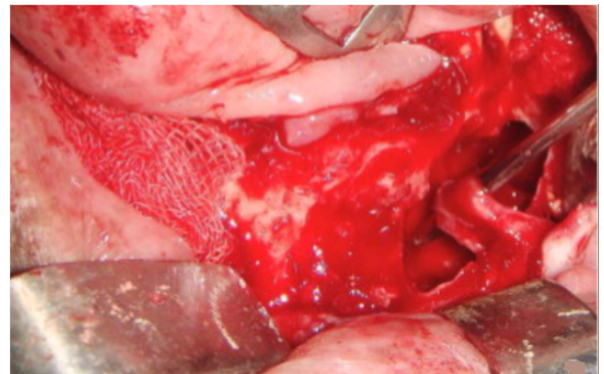
Computed tomography (CT) scan are suggestive of a cystic lesion in close proximity to the left maxillary sinus, and it appears to be associated with unerupted tooth structures. Here's a breakdown of the key observations: Unilocular Radiolucency, Well-Defined Borders, Radiopaque Tooth Structures. Given these CT findings, there is a strong suspicion that the lesion is consistent with a cystic condition, possibly a dentigerous cyst, associated with an unerupted tooth in the maxillary region.<sup>4</sup>



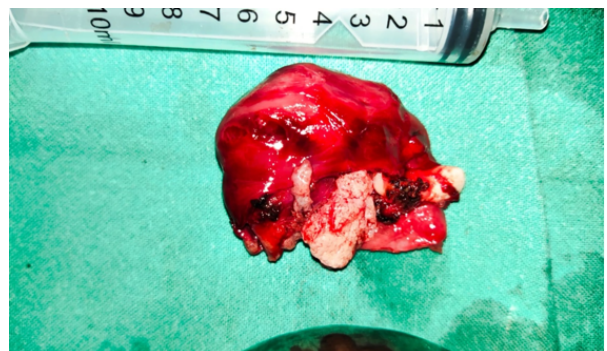
**Fig. 1:** (A) & 1(B): Swelling in the upper left side of the face(maxillary region)



**Fig. 2:** (A,B,C): CT Scan showing cyst like structure in left maxillary region



**Fig. 3:** Enucleation being done



**Fig. 4:** Removed specimen along with canine.

## 2.2. Treatment

Based on the clinical and radiological findings, the lesion was diagnosed as dentigerous cyst of the maxilla in association with unerupted tooth and surgical enucleation was planned under general anaesthesia. Caldwell luc operation and left medial maxillectomy with Denker's was done. Approx 20 ml fluid was aspirated before the incision to reduce the pressure. Abgel kept in the maxillary sinus cavity with 3 packs of merocel kept in the left nasal cavity and complete haemostasis achieved. Excised tissue sent for Histopathological examination.

## 2.3. Histopathology

The Specimen sent of excised cyst from left maxilla. The reports were suggestive of-Benign odontogenic tumour favouring Adenomatoid Odontogenic Tumour

## 2.4. Differential diagnosis

In this case, a provisional diagnosis of dentigerous cyst and mucocele of the maxilla was considered based on clinical examination. However, radiologically, several other differential diagnoses were also considered due to overlapping features.

**Keratocystic Odontogenic Tumor:** This is a type of odontogenic cyst that can have radiographic features similar to those of dangerous cysts.

**Inverted Papilloma with Secondary Mucocele:** Inverted papilloma is a benign tumor that can lead to the formation of secondary mucoceles in the nasal cavity.

## 2.5. Ameloblastoma

Ameloblastomas are benign tumors of odontogenic origin that can cause swelling and displacement of adjacent structures.<sup>5</sup>

## 3. Discussion

Adenomatoid Odontogenic Tumor (AOT) is considered a rare benign epithelial odontogenic tumor. Its prevalence can vary based on geographical location, and the relative frequency of AOTs can range widely, as reported in different studies. In a worldwide literature survey conducted by Philipsen et al., it was found that the relative frequency of AOTs could range from as low as 0.6% to as high as 38.5% in different regions.<sup>5</sup> This wide variation in reported frequencies may be due to several factors, including differences in population demographics, access to healthcare, and variations in diagnostic practices and reporting among different regions and healthcare systems. It's important for healthcare professionals to be aware of the potential existence of AOTs, even though they are relatively rare, to ensure accurate diagnosis and appropriate management when encountering such cases.<sup>6</sup> The summary

of distribution and features can be described as: Africa: Reported relative frequencies of AOTs in Africa range from 1% to 38.5%.Asia: Relative frequencies in Asia range from 1% to 16%.South America: AOTs are reported to account for 4-7% of odontogenic tumors in South America. North America: In North America, the relative frequency ranges from 2% to 7%. Middle East: In the Middle East, AOTs are reported to account for 2-4% of odontogenic tumors. Europe: The relative frequency of AOTs in Europe is reported to be in the range of 1-4%.Gender Predilection: AOTs are more commonly found in females, indicating a higher incidence in females than in males. Location: AOTs are often located in the anterior region of the maxilla. This location is notable for the presence of impacted teeth, and AOTs frequently develop around these impacted teeth. The maxillary anterior region includes the upper front teeth. It is indeed common for AOTs to be associated with an anterior segment of the maxilla and an impacted tooth, often involving the maxillary canine. This variation in the impacted tooth involved in AOT cases highlights the potential variability in the location of these tumors and their association with different teeth within the anterior segment of the maxilla. Nonetheless, the key characteristic is the presence of a cystic lesion associated with an impacted tooth in the anterior maxillary region, which aligns with the typical presentation of AOTs.As with any dental or maxillofacial condition, a thorough clinical examination, radiographic assessment, and histopathological examination are essential for accurate diagnosis and appropriate treatment planning in cases of AOTs. This helps ensure that the patient receives the most suitable care based on their specific presentation.<sup>6</sup> Maxillary canines are the most commonly affected teeth in cases related to the condition. In other words, in 51.3% of instances, this condition involves maxillary canines.<sup>7</sup> Histology also shows presence of capsule, necrosis, hyalinization, melanin pigmentation, and dysplastic dentinoid, osteodentin, calcification.<sup>8</sup>

However, in the case of dentigerous cysts, the impacted mandibular third molar is most commonly associated with this type of cyst. However, it's important to note that dentigerous cysts can also occur with other impacted teeth, including canines and premolars. Furthermore, the typical location of dentigerous cysts in the anterior maxilla or palate, which can result in swelling in the mid-palatal region. This swelling may extend into the maxillary antrum and nasal cavity, which can lead to clinical symptoms and complications. Due to their location and presentation, dentigerous cysts in the mid-palatal region should indeed be carefully differentiated from other causes of mid-palatal swellings, such as benign and malignant tumors, inflammatory conditions, or other cystic lesions. Given the potential variability in the impacted tooth involved and the range of clinical presentations, a comprehensive assessment,

including clinical examination, radiographic imaging, and possibly histopathological evaluation, is essential for an accurate diagnosis and appropriate management of dentigerous cysts to ensure the best possible outcome for the patient. The findings from our study align with common characteristics associated with dentigerous cysts. Dentigerous cysts are often slow-growing and can remain asymptomatic for a long period. The most common presenting symptom, as you mentioned, is often bony expansion. This means that as the cyst enlarges, it can cause the surrounding bone to expand or resorb, resulting in visible swelling or changes in the shape of the affected area<sup>4</sup>. Some surgeons have strongly argued in favor of the concept of AOT being derived from the complex system of dental lamina or its remnants and preferred the name AOT rather than adenomatoid odontogenic cyst (AOC).<sup>9</sup> AOT most of the times does not exceed 1-3 cm in diameter. Most cases of large AOTs described in the literature have been observed in the maxilla.<sup>10</sup>

#### 4. Conclusion

Our approach of performing a complete enucleation of the cyst is in line with common treatment strategies for managing dentigerous cysts and AOTs. While it's generally recommended to be as conservative as possible to minimize morbidity and preserve healthy tissue, it's also crucial to ensure that the cyst is entirely removed to prevent recurrence. Dentigerous cysts and tumors have the potential to recur if any part of the cyst lining or associated tissue is left behind during surgery. Therefore, complete enucleation, which involves the complete removal of the cyst or tumour and its lining, is often the preferred treatment approach. This helps minimize the risk of recurrence and ensures the best long-term outcome for the patient. Regular follow-up and monitoring after the surgery are also important to confirm that there is no recurrence and that healing is progressing as expected. Overall, the balance between being conservative to minimize morbidity and being thorough to prevent recurrence is a key consideration in the management. This case highlights the importance of regular ENT & dental check-ups, including radiographic examinations, to identify and manage dentigerous cysts in their early stages, before they lead to more serious complications or symptoms. Timely diagnosis and treatment are essential to ensure the best possible outcome for affected individuals.

#### 5. Consent for Participation

Taken.

#### 6. Availability of Data and Materials

Available in the manuscript in method section.

#### 7. Author's Contribution

1. SM Conceptualized, designed, has role in data collection, analysis, interpretation, surgical and medical practices, literature search and writing.
2. DB Conceptualized, designed, has role in data collection, analysis, interpretation, surgical and medical practices, literature search and writing.
3. SV Conceptualized, designed, has role in data collection, analysis, interpretation, surgical and medical practices, literature search and writing.

#### 8. Sources of Funding

No funds outsourced.

#### 9. Conflict of Interest

There are no competing or conflict of interest.

#### 10. Acknowledgment

Authors are highly thankful to Medical director Dr. V.K. Mahadik to carry out this case with all his support and encouragement.

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**Cite this article:** Mishra S, Bhojani D, Vaidya S. Adenomatoid odontogenic tumour of maxillary canine presenting as a dentigerous cyst: A case report. *IP J Otorhinolaryngol Allied Sci* 2023;6(3):85-89.