


# Adenomatoid Odontogenic Tumor Mimicking a Dentigerous Cyst in Maxilla

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## ABSTRACT

**Aim:** To present a case of adenomatoid odontogenic tumor (AOT) associated with impacted maxillary lateral incisor in a 12-year-old female that mimicked dentigerous cyst.

**Background:** Adenomatoid odontogenic tumor (AOT) was first mentioned by Steensland in 1905, which is a rare tumor of odontogenic origin. Dreibradt in 1907 coined the term pseudo ameloblastoma. In 1948, Stafne considered it a distinct pathological entity.

**Case description:** A 12-year-old female reported to the Department of Oral and Maxillofacial Surgery with the chief complaint of progressive swelling on the left maxillary anterior region for 6 months. The clinical and radiographical findings of the case represented a dentigerous cyst or unicystic ameloblastoma, but the histopathological interpretation was interpreted as AOT.

**Conclusion:** The AOT is an unusual entity that is commonly misdiagnosed as a dentigerous or odontogenic cyst. Histopathology plays a vital role in diagnosis and further management.

**Clinical significance:** The interest and relevance of the present case are the difficulties in diagnosing accurately based on the radiograph and histopathology. Both dentigerous cysts and AOT are entirely benign and encapsulated lesions, and enucleation poses no major difficulties. The case report highlights the importance of early diagnosis of neoplasm arising in odontogenic tissues. The fact that in cases of unilocular lesions surrounding the impacted tooth in the anterior maxillary region, AOT should also be considered as a differential diagnosis.

**Keywords:** Adenoameloblastoma, Adenomatoid odontogenic tumor, Dentigerous cyst, Teratomatous odontoma, Odontogenic tumor.

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## BACKGROUND

An AOT is an uncommon tumor of odontogenic origin, first described by Steensland in 1905.<sup>1</sup> Later, Dreibradt, in 1907, coined the term pseudo ameloblastoma. It is a slow-progressing and nonmalignant epithelial tumor. Clinical features of AOT—associated with impacted teeth or in absence, painless, bony expansion, facial asymmetry due to growth of the lesion.<sup>2</sup> This lesion is known by many names, including adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantium, or teratomatous odontoma.<sup>3–6</sup> The lesion was included in World Health Organization classification in 1971 based on Kalia et al. proposal, which was defined as “a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue.”<sup>2,3</sup> The AOT is of debatable origin, with a prevalence of 0.1% of cysts and tumors of the jaw and 3% of all odontogenic tumors. AOT has three variants—follicular, extrafollicular, and peripheral.<sup>4</sup>

## CASE DESCRIPTION

A 12-year-old female patient reported to our institution with left maxillary anterior swelling for 6 months. The patient complained of an increase in the size of the swelling and moderate pain in the past 2 weeks. On palpation, the lesion presented as diffuse, extraoral hard, non-tender 3 × 5 cm swelling in the anterior left maxillary region. Intraoral examination revealed a large compressible well-circumscribed swelling with a smooth surface in the buccal and palatal aspect of the left anterior maxilla (Fig. 1) with obliteration of the buccal vestibule. The patient had an over-retained left maxillary deciduous canine with a maxillary left lateral incisor and canine missing. There was a marked expansion of both the buccal and palatal cortices.

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Radiographic examination orthopantomogram (OPG) showed a large well-circumscribed unilocular radiolucency associated with the left anterior maxilla extending from the maxillary central incisor to the first molar region, with an embedded permanent lateral incisor within the lesion. There was a marked displacement of permanent canine towards the superior extent of the lesion. The cone-beam computed tomography (CBCT) revealed a lesion extending from the distal aspect of the left maxillary central incisor to the mesial aspect of the left maxillary first molar. Thinning of the palatal cortex was noted. The mesiobuccal root of the left maxillary first molar was partly resorbed. The left maxillary canine was pushed apically to the floor of the left maxillary sinus, which displaced the floor of the antrum and encroached upon the internal space of the antrum (Fig. 2).

Diagnostic aspiration fine needle aspiration cytology revealed straw-colored fluid. Based on clinical and radiographic findings, the differential diagnosis was of a dentigerous cyst or unicystic ameloblastoma.

After obtaining written consent, under general anesthesia, the cyst was completely enucleated along with the impacted left maxillary lateral incisor (Figs 3 and 4). The impacted left permanent maxillary canine, along with retained deciduous left maxillary canine, was extracted. The specimen was sent for histopathological examination, which revealed the characteristics of AOT that showed proliferation of epithelial cells in the form of ductal and rosette patterns, with evident cystic components. The ductal lumen was lined by an eosinophilic ring called a hyaline ring, and the ducts and rosette were surrounded by round and polygonal cells. Pale basophilic irregular to round calcified bodies was evident

between ductal and rosette pattern (Figs 5 and 6). Connective tissue showed collagen fibers interspersed with fibroblasts. The histopathological findings were consistent with AOT. Postoperative orthopantomogram shows there were no signs of recurrence on the 1-year follow-up of the patient (Fig. 7).

### DISCUSSION

In 1948, Stafne considered AOT as a distinct pathological entity.<sup>5</sup> This lesion is known by many names, including adenoameloblastoma, AOT, adamantinoma, epithelioma adamantium, or teratomatous odontoma.<sup>6</sup> The AOT is a slow-growing benign tumor representing 3% of odontogenic tumors. Philipsen et al.<sup>7</sup> study



Fig. 1: Intraoral view showing swelling on buccal aspect

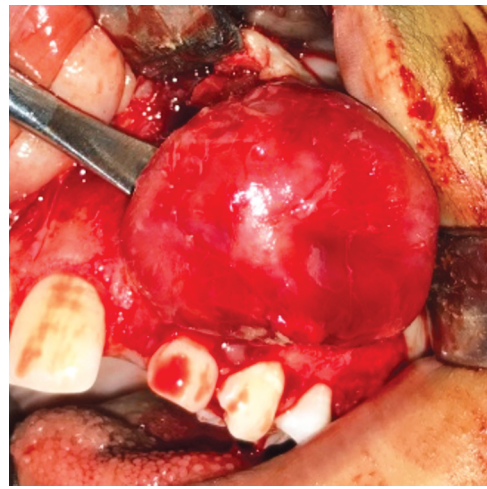


Fig. 3: Intraoperative image showing the lesion being enucleated

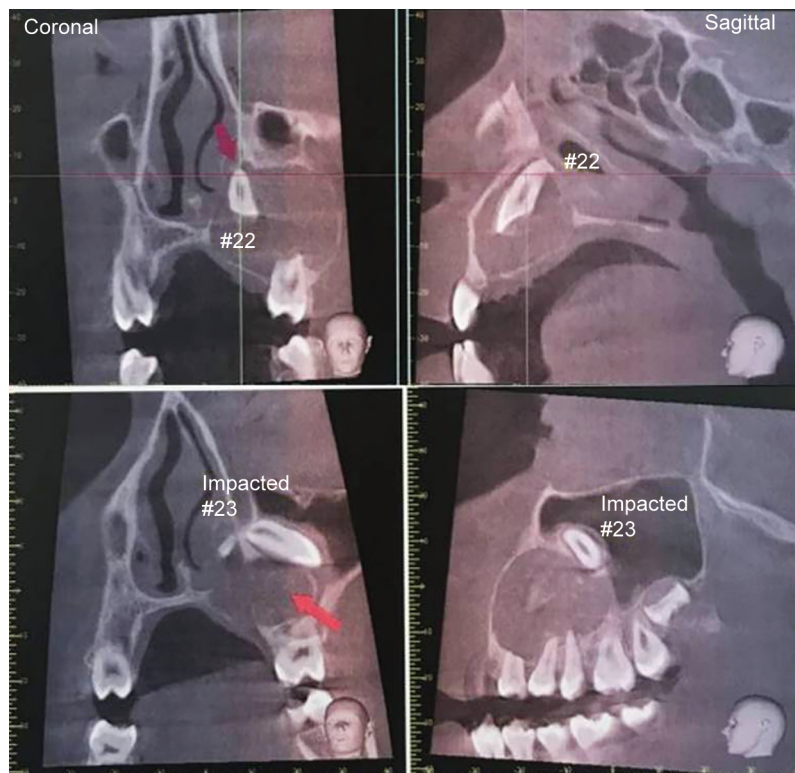
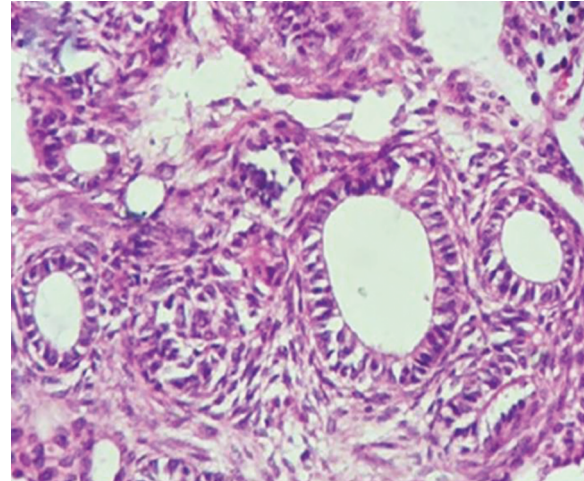


Fig. 2: CBCT images showing impacted maxillary left lateral incisor with well-circumscribed radiolucency and an impacted left maxillary canine

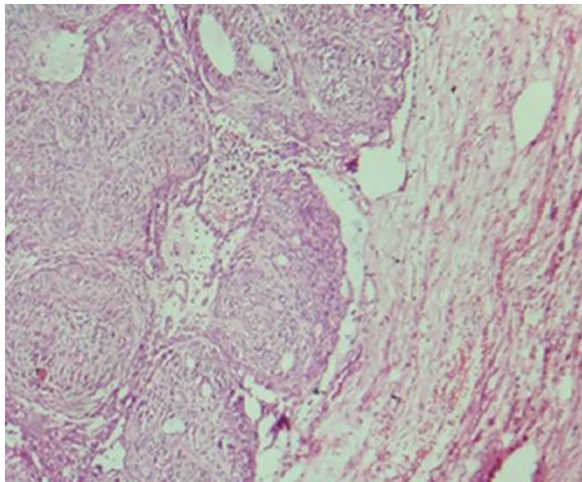




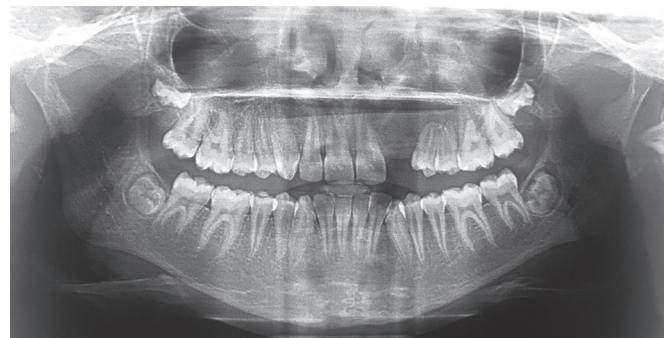
**Fig. 4:** Specimen showing the enucleated lesion with impacted maxillary left lateral incisor



**Fig. 6:** Tumor cells arranged in duct-like patterns and rosettes



**Fig. 5:** Histopathologic photomicrograph reveals fibrous sheath encapsulating islands of tumor cells



**Fig. 7:** Postoperative OPG

based on 39 cases revealed the prevalence of AOT two-thirds in the second decade of life and over 50% in the age distribution 13–19 years.<sup>7</sup> The tumor presents as a partially cystic or solid lesion in the wall of the large cyst, often misdiagnosed as a dentigerous cyst.<sup>8</sup> The follicular and extrafollicular variants have a 96% prevalence, of which the follicular variant has a 71% occurrence. The follicular variant is predominantly associated with the crown and is often part of the root of an impacted (unerupted) tooth. The most frequently associated tooth is the maxillary canine, rarely the permanent molars. The peripheral variant is very rare, with only 18 cases reported so far in the literature.<sup>3</sup>

The incidence of AOT is higher in females and is usually located in the anterior region of the maxilla within which an impacted tooth exists.<sup>2</sup> Many cases have been reported that the AOT is associated with an anterior segment of the maxilla along with an impacted canine. The case we present here is also similar, except that the tumor is associated with an impacted permanent lateral incisor.

The origin of AOT is hypothesized to be from the odontogenic epithelium of the dentigerous cyst, while some believe that the tumor could be derived from epithelial remnants of the dental lamina complex system, making it controversial. Suppose the lesion grows into a nearby dental follicle or next to the follicle leading to

the “developmental” theory. If AOT is derived from a dentigerous cyst, it is suggestive of a hybrid variant as suggested by Chen et al.<sup>9</sup> In our case; the tumor surrounded the fully formed impacted maxillary lateral incisor suggesting developmental pathogenesis or “hybrid variant.”<sup>10</sup>

The present case was challenging as it was difficult to diagnose based on the radiographic and histopathological findings. Both dentigerous cysts and AOT are entirely benign and encapsulated lesions, and enucleation poses no major difficulties. The involved teeth can be saved, considering the age of the patient, if the dental follicle is found to be uninvolved and can be easily separated from the tumor. In the present case, permanent canine in close approximation to the lesion made it impossible to save the tooth. No aggressive behavior on the part of the adenomatoid tumors has been described, and recurrence is very rare following complete enucleation of the primary lesion.

## CONCLUSION

In this case, it was difficult to make a final diagnosis without a microscopic examination. The clinical and radiographic findings were used to get a differential diagnosis, but those findings were not enough for the final diagnosis. The histopathologic finding

showed the proliferation of epithelial cells in the form of ductal and rosette patterns, which is a characteristic feature of the AOT. The AOT is a rare, benign, and slow progressive jaw lesion that is commonly misdiagnosed as a dentigerous cyst. Histopathological interpretation plays a vital role in the diagnosis of AOT.

### Clinical Significance

The present case report highlights the importance of early diagnosis in rapidly growing swellings in the anterior maxillary region, especially in young female patients, as it could be AOT which will require aggressive surgical intervention. Swelling associated with over-retained deciduous teeth and unerupted permanent teeth must be investigated on a priority basis to rule out any odontogenic pathology like AOT. Early diagnosis will lead to less postoperative morbidity, which is of paramount importance as the lesion is mostly associated with the anterior maxillary region, which is of high esthetic concern for young adolescent females.

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