



CASE REPORT

Adenomatoid odontogenic tumor mimicking a dentigerous cyst

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Received 24 January 2005; accepted 26 March 2005

KEYWORDS

Adenomatoid odontogenic tumor;
Odontogenic cyst;
Odontogenic tumor

Summary Adenomatoid odontogenic tumor (AOT) is a slow-growing, asymptomatic and uncommon lesion that arises from odontogenic epithelium with inductive effects on connective tissue. The more common variant is the follicular type, which involves an unerupted tooth and is often mistaken for a dentigerous cyst. Histopathologic examination demonstrates cuboidal or spindle-shaped epithelial cells forming aggregates or rosette-like structures with minimal connective tissue, and cuboidal or low columnar cells forming glandular duct-like structures. Treatment is complete enucleation, and recurrences are rare. We presented a case of AOT in a 14-year-old male presenting as a cystic mass around an unerupted tooth.

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1. Introduction

Adenomatoid odontogenic tumor (AOT) is relatively rare, and was previously described as a histological variant of ameloblastoma but is now recognized as a separate entity by the World Health Organization [1]. It is a slow growing lesion that is thought to arise from odontogenic epithelium because of its predilection for tooth-bearing bone, with varying degrees of inductive change in connective tissue. It often causes expansion of surrounding bone and displacement of adjacent teeth. The most common presentation is a cystic mass involving an unerupted tooth.

The exact incidence or prevalence of AOT is unknown. The available pathological data on odontogenic tumors does give the relative frequency of

AOT at 2.4–6.8% [1–3]. Most reviews tend to agree on a slight female predominance of 1:1.2–2 [1,4–6]. Because of the slow growing and generally asymptomatic nature of the lesion most patients would tolerate the mass for years until it has produced a significant or obvious deformity and discomfort. Typically, the tumor would be 3–4 cm in maximum diameter at the time of clinical presentation [7–9].

2. Case report

A 14-year-old boy presented at the Cincinnati Children's Hospital Medical Center with a firm swelling in the left cheek. The occurrence of sharp pain around the area over a period of three weeks prompted the consult. Clinical examination

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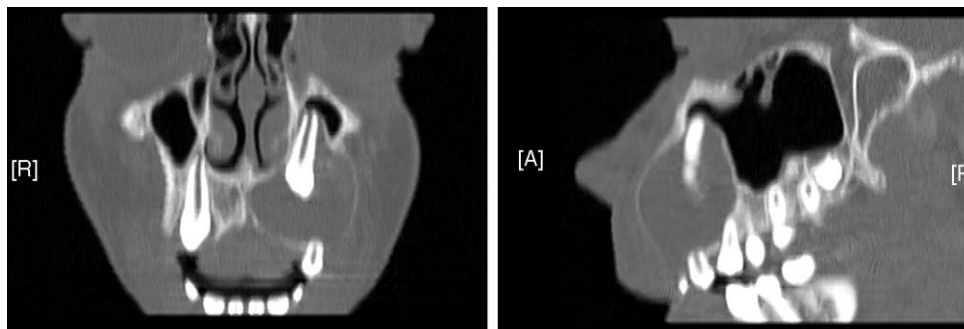


Fig. 1 Pre-operative non-contrast enhanced CT taken in coronal and sagittal views demonstrate cystic nature of lesion, unerupted tooth and local expansive changes.

revealed a smooth bulge in the left upper alveolar ridge extending onto the anterior maxillary wall, absence of the left upper canine and mild displacement of the adjacent lateral incisor and first premolar. Preoperative CT scans demonstrated an expansile lesion with a sclerotic rim in the left anterior maxilla involving an unerupted tooth (Fig. 1). The lesion extended into the maxillary sinus and displaced the adjacent lateral incisor and first premolar. It measured 3.4 cm × 3.0 cm × 2.8 cm with no contrast enhancement, and this was interpreted as a dentigerous cyst.

The mass was enucleated completely under general anesthesia through a Caldwell–Luc approach. A small window was made into the cyst, and brownish gelatinous fluid was obtained. The involved tooth was visualized within its lumen, surrounded by areas of thickening of the cyst lining. The cyst separated easily from the adjoining bone, and was removed

with the involved tooth (Fig. 2). The surrounding bone was polished with a diamond burr.

3. Histopathological examination

The surgical specimen consists of a cyst that measures 3.4 cm in maximum diameter surrounding the crown of a grossly unremarkable tooth. The outer surface of the cyst is smooth. The cyst wall measures 0.5 cm in maximum thickness in most areas. However, the cyst cavity is partially filled by white/pink solid tissue with a fleshy cut surface.

Sections of the solid tissue show a tumor with a rim of fibrous pseudocapsule. The tumor has a nodular growth pattern and is composed of a dual cell population (Fig. 3A). One cell population is composed of spindle-shaped and polyhedral cells that form large aggregates (Fig. 3B). These cells do not

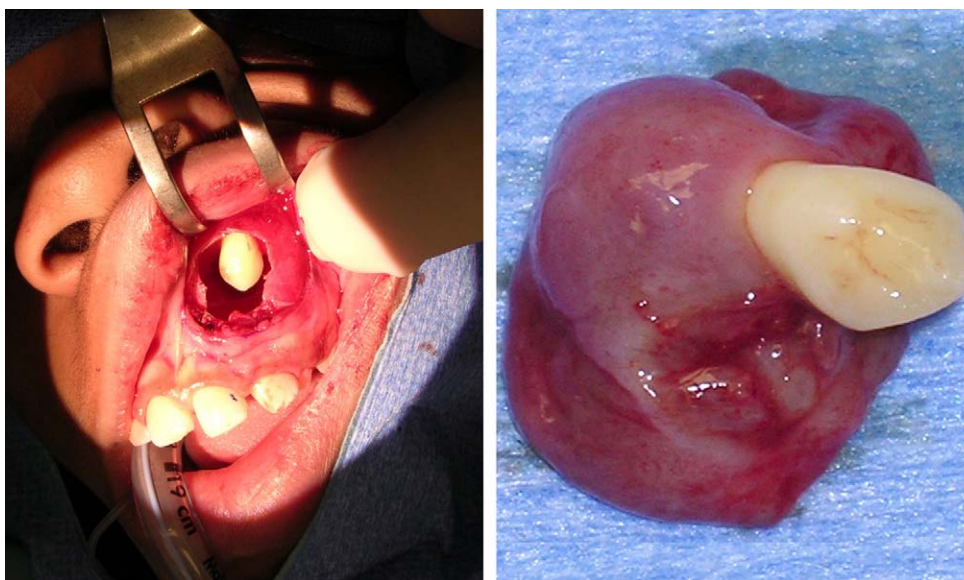


Fig. 2 Intraoperative photographs. A window has been created on the anterior surface of the cyst, and the involved canine is clearly visible. The cyst lining is dissected off the surrounding bone, and the tooth is easily removed with the cyst. The cyst has been turned inside out to show inner lining and tooth.

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