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## Case Report

# Adenomatoid odontogenic tumor of the mandible with a large calcified mass: Report of a rare case

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## ABSTRACT

Adenomatoid odontogenic tumors (AOTs), relatively uncommon benign lesions of odontogenic epithelial origin, are categorized into three clinicopathological variants: follicular, extrafollicular, and peripheral. We present a rare case of follicular AOT with an impacted tooth and a large calcified mass. A 13-year-old girl was referred by her orthodontist for evaluation of the delayed eruption of a left mandibular lateral incisor and a radiopaque mass of the anterior mandible. Computed tomography showed an odontoma-like radiopaque mass adjacent to the crown of the impacted left mandibular lateral incisor inside the radiolucent area. The lesion was enucleated totally as one mass with the unerupted tooth under general anesthesia. The histopathologic findings, such as the cuboidal or columnar epithelial cells forming duct-like or rosette-like structures and intercellular droplets of amorphous eosinophilic material, corroborated the AOT characteristics reported in the literature. However, the large calcified mass was a rare finding in follicular AOT. The current case of AOT was difficult to distinguish from the more common forms of odontogenic lesions. There have been no signs of recurrence of the AOT 14 months postoperatively.

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## 1. Introduction

Adenomatoid odontogenic tumors (AOTs) are benign, asymptomatic, uncommon odontogenic lesions that account for 2–7% of all odontogenic tumors [1–3]. In 1907, Dreidblatn first described AOT as an adenoameloblastoma. AOTs have been described since the early 1900s under various names, including cystic adamantoma, adenoameloblastoma, cystic complex composite odontoma, adenoameloblastic odontoma, ameloblastic adenomatoid tumors, and ameloblastic odontogenic tumors. In 1948, Stafne first recognized AOT as a distinct entity, but many authors have classified it as a variant of an ameloblastoma [1,4]. Philipsen and Birn proposed the widely accepted and currently used name, AOT, in 1969, which the World Health Organization (WHO) adopted in 1971 in its first edition of the classification of odontogenic tumors [5,6]. AOT generally is regarded as a true neoplasm of odontogenic origin.

AOT, which has a female predominance, occurs most often during the second and third decades of life. The maxilla is the preferred site, almost twice as often as the mandible, and the anterior region is affected more frequently than the posterior area [2,6–11]. The lesion is characterized by slow but aggressive growth that causes a painless expansion of the jaw and often is associated with impacted teeth, frequently canines or lateral incisors [2,8–11]. AOT is often discovered incidentally during a routine radiographic examination for dental treatments.

Most cases of AOT have areas of small calcified material [1,7]. To our knowledge, few reports have been published on AOTs associated with a large calcified mass. We present a rare case of AOT associated with an impacted mandibular lateral incisor and a large calcified mass in a 13-year-old girl.

## 2. Case report

A 13-year-old girl was referred by her orthodontist for evaluation of the delayed eruption of a left mandibular lateral incisor and a radiopaque mass of the anterior mandible. There was no relevant medical history and the patient was in good general health. No swelling of the anterior mandibular gingiva was seen intraorally (Fig. 1). A panoramic radiograph revealed a well-circumscribed, unilocular, round radiolucency involving the impacted left mandibular lateral incisor (Fig. 2). Inside this radiolucent area, a radiopaque mass was adjacent to the crown of the

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**Fig. 1.** An intraoral view shows no swelling of the anterior mandibular gingiva.



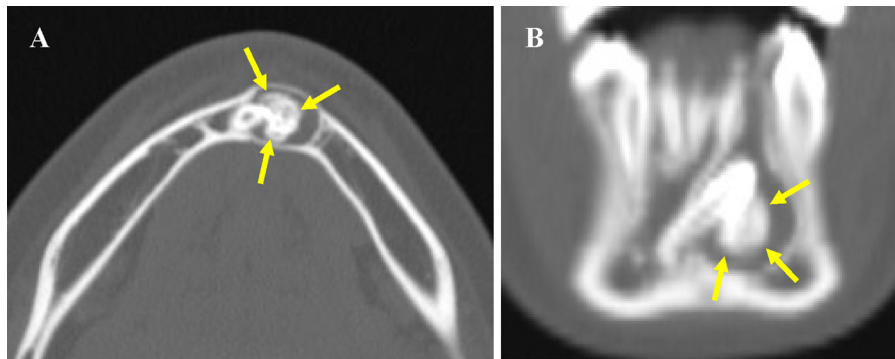
**Fig. 2.** A well-circumscribed, unilocular, round radiolucency involving the impacted left mandibular lateral incisor and a radiopaque mass adjacent to the crown of the impacted tooth are seen in the panoramic radiographic image.

impacted tooth. A computed tomography (CT) scan showed slight expansion of the labial alveolar bone (Fig. 3). The labial cortical plate was very thin, with no complete resorption. The size of the radiopaque mass was about 11 mm × 11 mm in diameter. Based on these radiographic findings, the current case was diagnosed as a cystic complex odontoma.

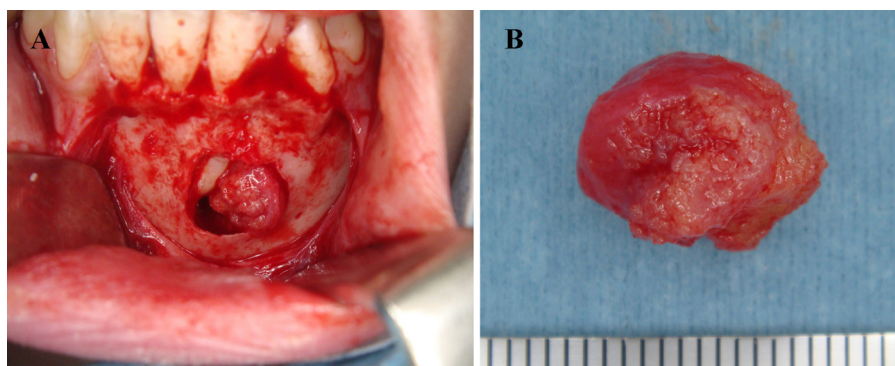
Intraoperatively, the lesion was enucleated as one mass with the impacted tooth under general anesthesia (Fig. 4A). Histopathology of the surgical specimen showed a firm lesion about 15 mm in maximal diameter (Fig. 4B). Microscopy showed a well-defined cystic tumor surrounded by a thick fibrous connective tissue capsule (Fig. 5A and B). The tumor was comprised of nodules of varying sizes consisting of cuboidal or columnar epithelial cells forming duct-like or rosette-like structures (Fig. 5B). Duct-like structures were lined by one or two layers of columnar epithelial cells with the polarized nuclei away from the luminal surface (Fig. 5C). Inter-cellular droplets of amorphous eosinophilic material also were seen in the rosette-like areas (Fig. 5D). The characteristic calcified mass was adjacent to the tumor (Fig. 5A). We needed to decalcify the specimen because the calcified mass was too large. The areas of eosinophilic calcification were observed (Fig. 5E). The features were compatible with a diagnosis of AOT. The patient has been followed for 14 months without a recurrence.

### 3. Discussion

AOT, an infrequently occurring benign lesion of odontogenic epithelial origin, is mostly encountered in young patients, especially in the second decade of life, and develops more often in females than in males [2,6–11]. The female predilection is even more common in Asian females, with the highest prevalence observed in Sri Lanka (3.2:1) and Japan (3:1) [12]. The



**Fig. 3.** A preoperative CT scan demonstrates slight expansion of the labial alveolar bone and a radiopaque mass inside radiolucent area (arrows) on (A) an axial section and (B) a coronal section.



**Fig. 4.** (A) An intraoperative photograph shows that the labial cortical plate on the surface of the lesion, and the lesion, and the involved lateral incisor has been excised. (B) A photograph of the surgical specimen.

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