

## Unicyclic ameloblastoma of the maxilla : A case report

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**ABSTRACT** A 26-year-old male was referred to our hospital complaining of an asymptomatic swelling of the left posterior maxilla. The patient's dental history indicated that his left maxillary third molar had been extracted approximately three years previously. At presentation, radiography demonstrated a well-defined 3.5 cm diameter radiolucency of the left maxilla. The lesion was enucleated and histopathological examination confirmed a diagnosis of unicyclic ameloblastoma, plexiform type (type 3b).

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Key words : Ameloblastoma , Unicyclic , Maxilla , Enucleation

### INTRODUCTION

Ameloblastoma is a benign odontogenic tumor that can be locally aggressive and invasive. Unicyclic ameloblastomas (UAs) account for 6 to 15% of all intraosseous ameloblastomas in various studies and account for more than 90% of those found in the mandible<sup>1,2)</sup>. We report a rare case of UA of the maxilla.

### CASE REPORT

A 26-year-old male with a fistula of the left maxillary region was referred to our department from an outside dental clinic on September 16, 2006. His dental history indicated that the left maxillary third molar had been extracted for pericoronitis at another hospital in 2003. However, there had been incomplete healing of the extraction socket. After that, he moved and there was no further intervention

until his recent presentation.

Intraoral examination revealed a fistula at the gingivobuccal fold of the left maxillary second and third molars, and there was a soft mass, the size of an egg, under the fistula (Fig. 1). Panoramic radiography disclosed an oval-shaped radiolucent area with well-defined limits of 3.5 cm diameter



Fig. 1. Intraoral examination showing a fistula at gingivobuccal fold of the left maxillary second and third molars, and there is a soft mass, the size of an egg under the fistula.

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radiolucency of the left maxilla (Fig. 2). CT images revealed that the tumor filled the left maxilla and was separated by a septum in the maxillary sinus (Fig. 3a, b). Although an incisional biopsy was performed, the diagnosis was granulation tissue. Fine needle aspiration was carried out and revealed ameloblastoma cells. Enucleation of the lesion, left maxillary second molar extraction, and a Caldwell-Luc operation in the left maxillary sinus were carried out under general anesthesia. The lesion adhered to adjacent bone. The size of the lesion was  $40 \times 30 \times 12$  mm and it had a thick and grayish-white coloration. Histological examination revealed a part of the unicystic tumor consisting of plexiform structures of proliferating cords and strands of odontogenic epithelial cells. The peripheral part of the tumor cord showed columnar cells with oval nuclei with their polarization away from the basement membrane (Fig. 4a, b). The histopathological diagnosis was a UA, plexiform type (type 3b).

There have been no signs of recurrence during the 27-month follow-up period.

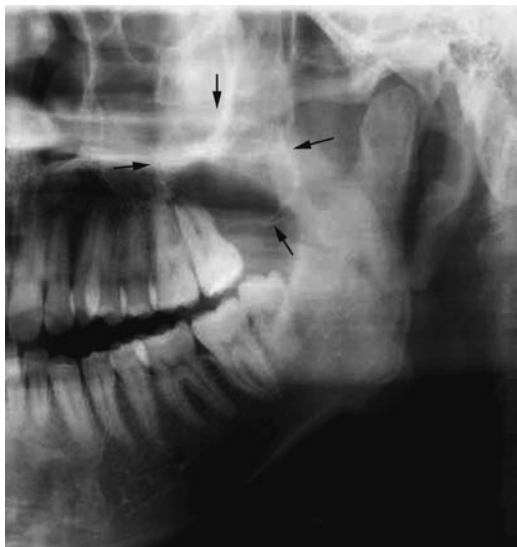


Fig. 2. Panoramic radiography showing a large unilocular radiolucent lesion involving the mandibular body in the edentulous first and second molar periapical areas (arrows).

## DISCUSSION

UA was first described as a distinct variant of ameloblastoma in 1977 by Robinson and Martinez<sup>3)</sup>. Thereafter, Ackermann *et al.*<sup>1)</sup> reclassified UA into three types with prognostic and therapeutic implications. In type 1, the tumor is confined to luminal surface of cyst with a lining of ameloblastomatous epithelium. Type 2 is characterized by epithelial nodules arising from the cystic lining and projecting into the cyst lumen. There is no evidence of infiltration of the fibrous cystic wall in either type of lesion. In type 3, the fibrous wall of cyst is infiltrated by a trabecular



Fig. 3. (a) Axial CT scan showing the lesion expansion of the left maxillary sinus. (b) Coronal CT scan showing the lesion in the maxilla (white arrow) and opacification of the left maxillary sinus (black arrow).

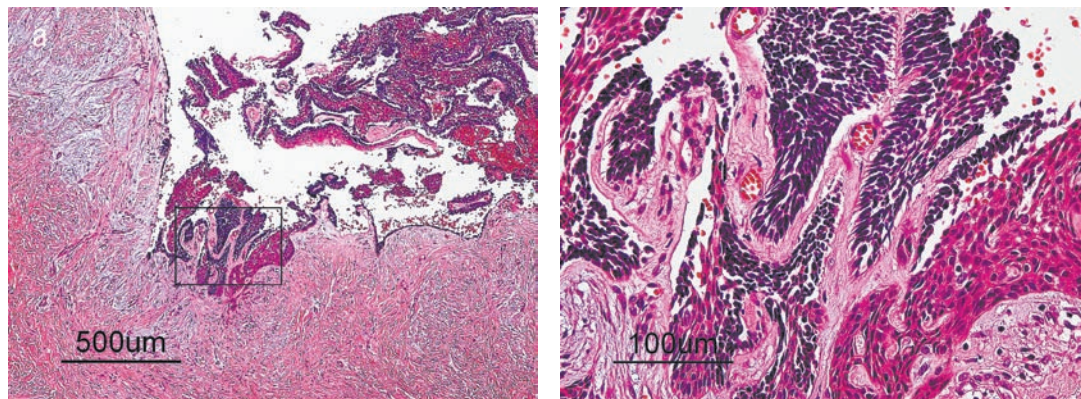


Fig. 4. (a) Histopathology showing plexiform structure of proliferative cords and strands of odontogenic epithelial cells (hematoxylin-eosin stain). (b) Odontogenic epithelium having hyper-chromatic polarized basal layer; intramural nodule with focal invasion of ameloblastoma (hematoxylin-eosin stain).

pattern that resembles the plexiform pattern seen in conventional ameloblastoma. In addition, it is characterized by a basal layer of columnar cells with hyperchromatic nuclei. These cells are loosely cohesive and resemble stellate reticulum epithelium.

The radiographic features of UA are typically unilocular and there is a round area of radiolucency. Therefore, this lesion is often misdiagnosed as an odontogenic keratocyst or a dentigerous cyst<sup>4)</sup>. Konouchi *et al.*<sup>4)</sup> performed contrast enhanced (CE)-MRI to diagnose 13 cases of unilocular, round radiolucent lesions visualized by panoramic radiography and/or CT. In the cases of UA, low signal intensity (SI) was observed on T1-weighted images (WIs), and a markedly high SI was observed on T2WIs ; and relatively thick rim-enhancement with/without small intralimbal nodules was observed on CE-T1WIs. CE-MRI was considered to be useful in the diagnosis of UA.

Various treatment modalities for UA have been used, such as segmental or marginal resection as normally used for conventional ameloblastoma. However, more conservative treatments have been reported frequently, including enucleation, curettage, and marsupialization<sup>5, 6)</sup>. A conservative approach to treatment is important in the age group affected with the unicystic variant because

in a younger person the growth of the jaws is not yet complete. Akasaka *et al.*<sup>7)</sup> investigated the immunohistochemical discrepancy between UAs and other types. Expression of proliferating cell nuclear antigen (PCNA) was markedly observed in the tumor cells of other types of ameloblastoma, whereas there was no expression of PCNA in the cells of any variants of UA. Moreover,  $\beta$ -catenin was characterized by a more positive marked expression in the UA than in other types of ameloblastoma, and the cells that expressed this substance were not PCNA positive cells. For the reasons mentioned above, UA should be treated with a conservative therapy. In the present case, we selected enucleation and the patient was able to avoid facial deformity and oral dysfunction.

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