CASE REPORT

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# Surgical Treatment for a Huge Maxillary Ameloblastoma via Le Fort I Osteotomy: A Case Report

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Ameloblastomaa are odontogenic benign tumors with epithelial origin, which are characterized by slow, aggressive, and invasive growth. Most ameloblastomas occur in the mandible, and their prevalence in the maxilla is low. A 27-year-old male visited our clinic with a chief complaint of the left side nasal airway obstruction. Three-dimensional computed tomography showed left maxillary sinus filled with a mass. Except for the perforated maxillary left edentulous area, no invaded or destructed bone was noted. The tumor was excised via Le Fort I osteotomy. The main mass was then sent for biopsy and it revealed acanthomatous ameloblastoma. The lesion in the left maxillary sinus reached the ethmoidal sinus through the nasal cavity but did not invade the orbit and skull base. The tumor was accessed through a Le Fort I downfracture in consideration of the growth pattern and range of invasion. The operation site healed without aesthetic appearances and functional impairments. However, further long-term clinical observation is necessary in the future for the recurrence of ameloblastoma. Conservative surgical treatment could be the first choice considering fast recovery after surgery and the patient's life quality.

Key Words: Acanthomatous ameloblastoma; Le Fort I downfracture; Maxillary sinus; Tumor

## Introduction

Ameloblastomas are slow-growing benign tumors that account for 1% of all tumors of the jaws[1)]. These tumors originate from the epithelial tissue involved in tooth formation[2)]. According

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to the 2017 World Health Organization (WHO) histological classification of head and neck tumors, ameloblastomas are classified into three subtypes: unicystic, extraosseous/peripheral, and metastasizing ameloblastomas[3]. On the basis of discernible histological patterns, the tumors can be categorized into follicular, plexiform, acanthomatous, granular cell, basal cell, and desmoplastic types. Among these, the most prevalent type is the follicular variant (64.9%), whereas the acanthomatous variant (3.9%) is the rarest[1,4,5].

Ameloblastomas mainly arise from the mandible in patients in their 30s and 40s. The reported incidence of ameloblastomas arising from the maxilla and mandible varies from 1:8.8 to 1:58[6]. Because of the characteristics such as local invasive growth and high recurrence rate, accurate histological diagnosis and an appropriate surgical approach are required for the management of these tumors. Reported recurrence rate for the conservative treatment is around 40% and for the radical treatment, around 12%. Conservative approaches such as marsupialization and subsequent marginal resection is an adequate option in maxillary cases to preserve the integrity of the bones and other anatomical structures for patients aged less than 20 years[7].

This case report presents a conservative surgical treatment for a huge maxillary ameloblastoma extending from the skull base to the alveolar bone of the maxilla with a rare histological subtype.

## Case Report

A 27-year-old male patient with glomerulonephritis presented to our hospital with a complaint of nasal airway obstruction. He had no other medical history. He had undergone sinus surgery at the Oral and Maxillofacial Surgery department in 2014 and the Ear, Nose, and Throat (ENT) department of another hospital in 2017 for the same issue. At ENT, surgery with incisional biopsy revealed a pilomatrixoma presenting as a sinus mass.

The patient had no facial deformities. From a three-dimensional computed tomography (CT), slight

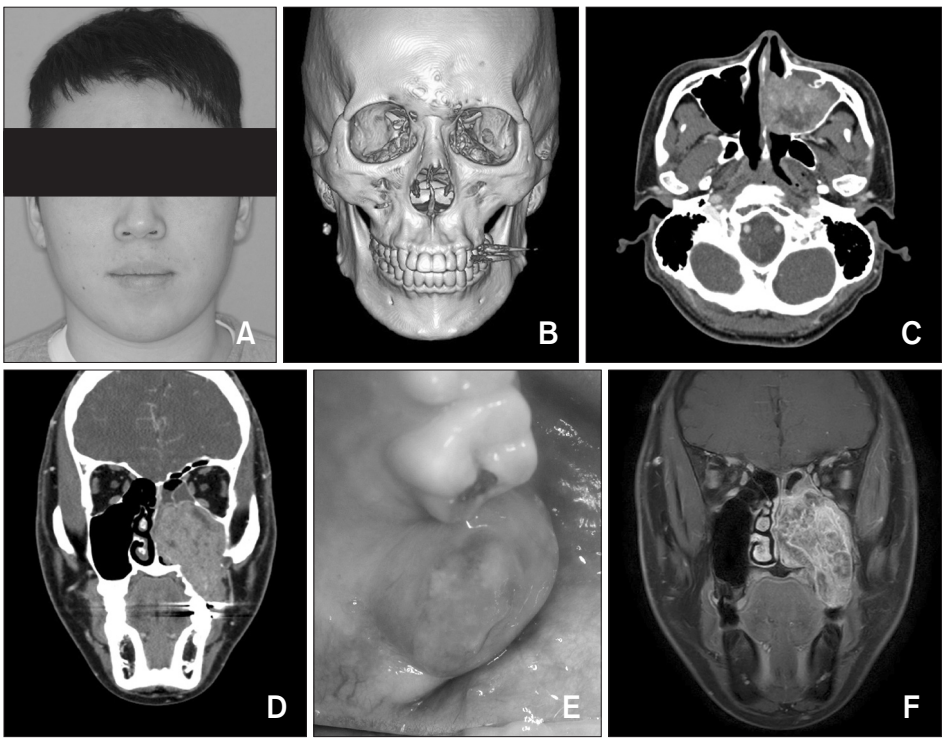


**Fig. 1.** (A) Patient's facial photograph. No significant facial deformities were present. (B) Three-dimensional computed tomography (CT) view. (C) Axial view in enhanced CT. (D) Coronal view in enhanced CT. (E) The tumor perforated the left second molar area mucosa and protruded into the oral cavity. (F) Coronal view in magnetic resonance imaging showed mucosal thickening at the patient's left ethmoidal sinus and the thickened mucosal margin had a boundary with the mass.

expansion of the maxillary sinus wall was found. No destructed bone except for the perforated alveolar bone and gingiva in the missing maxillary left second molar area was seen. The CT examinations showed that the mass had filled the left sinus from the posterior part of the maxillary alveolar bone to the skull base. Magnetic resonance imaging (MRI) showed about 5.2 cm×4.6 cm×5.8 cm sized, relatively well-defined lobulating mass in the left maxillary sinus area with expansive growth pattern and mucosal thickening in the left ethmoidal sinus (Fig. 1).

The selected surgical approach involved excision via Le Fort I osteotomy because of the quality of life of the patient. This surgical approach could minimize damage to around anatomic structures. The mass protruding into the mouth with a diameter of around 1 cm was dissected from the oral mucosa and the nasal mucosa. After removal of the mass in the nasal cavity, mega-antrostomy involved turbinectomy was performed to prevent recurrence and to secure the nasal airway. The mass was thus excised completely and was sent for biopsy with the mucosal segments of the nasal cavity. The maxilla was repositioned and fixed with plate-and-screw fixation. The oro-antral fistula in the left maxillary posterior edentulous area was about 1.5 cm in diameter and the fistula was closed with a pedicled buccal fat pad. The tumor was excised as several pieces and the biggest mass was about 4.5 cm×4 cm×2.5 cm in volume (Fig. 2). The sinus membrane was clearly removed, and the nasal mucosa was sent for the frozen biopsy. The biopsy result was negative. The patient showed rapid recovery after surgical treatment and was discharged within a week after uneventful recovering.

The biopsy findings revealed that the mass was an acanthomatous ameloblastoma, which was quite different from the previous biopsy result. Histological study showed that the mass had reverse polarity, which is a characteristic of the ameloblastoma, and showed no calcification, a characteristic of a pilomatrixoma. After surgery, the patient's chief complaint was resolved and showed no recurrence or abnormal signs over a 6-month follow-up period.

## Discussion

Ameloblastomas are benign tumors that show local invasive growth and a high recurrence rate[6]. Most ameloblastomas occur in the mandible, and

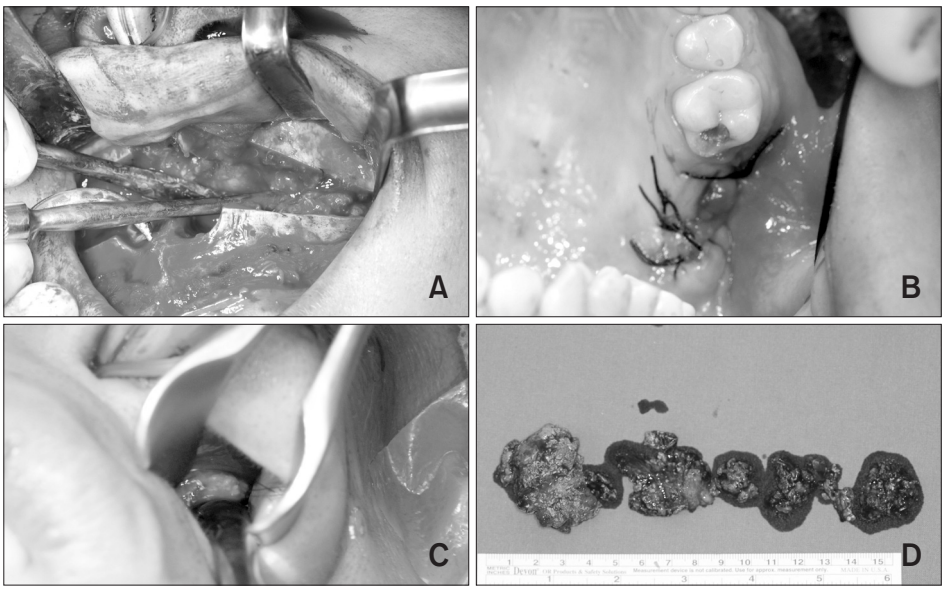


**Fig. 2.** (A) Intraoperative clinical photograph. Lesion was excised via the Le Fort I osteotomy approach. (B) Perforated gingival mucosa covered with a pedicled buccal fat pad. (C) The tumor also obstructed the nasal airway. (D) Excised tumor. The mass was about 4.5 cm×4 cm×2.5 cm in size.

radiological studies are helpful to diagnose tumors that occur within the bones. However, these tumors can occur in the maxillary sinus and grow to the skull base without causing bony deformities, making radiological diagnosis difficult. Thus, in addition to radiological examinations, biopsy can be performed to make a correct diagnosis and to select an appropriate treatment option.

In this case, previous biopsy revealed that the mass was a pilomatrixoma. Pilomatrixomas are firm and well-defined benign tumors derived from the outer sheath cell of the hair follicle root[8]. These tumors usually occur in the head and neck region in patients aged 20 years and older and have a female:male incidence ratio of 3:2[9]. Similar to acanthomatous ameloblastomas, pilomatrixomas are uncommon tumors, but their clinical characteristics and prognosis are quite different from those of ameloblastomas. Therefore, histological data should be combined with clinical findings for an accurate diagnosis. Unlike ameloblastomas, pilomatrixomas have a low recurrence rate and show good prognosis after surgical removal[10,11].

According to the WHO histological classification of head and neck tumors in 2017, ameloblastomas can be classified into three subtypes: unicystic, extraosseous/peripheral, and metastasizing ameloblastomas[3]. On the basis of discernible histological patterns, ameloblastomas can be also categorized as follicular, plexiform, acanthomatous, granular cell, basal cell, and desmoplastic. The follicular variant is the most prevalent type (64.9%), whereas the acanthomatous variant (3.9%) is the most rare type[1,4]. Regardless of the histological subtype, conventional ameloblastomas show similar prognosis because of their characteristically high recurrence rate and local invasive growth. The unicystic type shows a lower recurrence rate

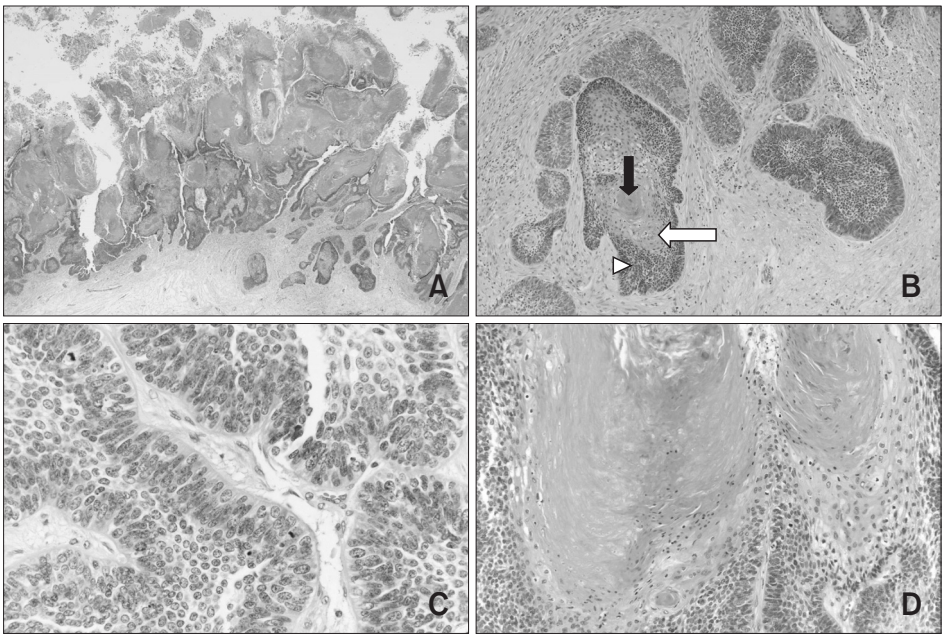


**Fig. 3.** Histological findings. (A) H&E staining (×20). At low magnification, most of lining epithelium showed marked keratinization in the center and upper portions of the epithelial islands. (B) H&E staining (×40). The tumor is composed of characteristic epithelium of the ameloblastoma in the fibrous stroma. The basal cells of these islands (arrowhead) are columnar and hyperchromatic and show a palisading pattern. The central part of the islands (black arrow) shows squamous differentiation. Marked keratinization is noted in this area (white arrow). (C) H&E staining (×400). Palisaded cells and Columnar cells have a tendency for the nucleus to move from the basement membrane to the opposing end of the cell, a process referred to as reverse polarization. (D) H&E staining (×100). Squamous differentiated cells are benign. Marked keratinization with parakeratosis is noted.

than conventional ameloblastoma[7]. In this case, biopsy proved that the mass was an acanthomatous ameloblastoma, a rare histological subtype. The tumor was composed of the characteristic epithelium of ameloblastomas in fibrous stroma. The basal cells of these islands were columnar and hyperchromatic and showed a palisading pattern. The central part of the islands showed squamous differentiation. Marked keratinization was noted in these areas (Fig. 3).

Because of the high recurrence rates and local invasive growth patterns, resection with a safe margin and immediate reconstruction is the usual treatment approach for these tumors. Resection and reconstruction are relatively easier to perform in mandible surgery because of the absence of nearby vital structures. However, in the maxilla, conservative approaches such as marsupialization, excision, and curettage could be better options considering the anatomical structure and the patient's quality of life. Following the conservative treatment, a long-term follow-up is essential because most ameloblastomas are knowm to recur within five years. Follow-up assessments should include panoramic radiography performed at 6-month intervals over the first 5 years and once every year until 10 years. After 10 years, follow-up assessments once every 2 to 3 years should be performed for as long as possible[7].

In this case, there were three possible approaches; segmental osteotomy, Caldwell-Luc approach, and the Le Fort I osteotomy. The left segmental osteotomy was too risky and challenging because of the possible teeth injury. Moreover, the tumor was too huge to remove using Caldwell-Luc approach. Therefore the patient underwent excision via Le Fort I osteotomy. This approach is a safe procedure with a low incidence of the complications. The major complications of Le Fort I osteotomy may be anatomical complications. Kramer et al.[12] reported that the nasal septal deviation was 1.6% and malposition of the maxilla was 0.2% of the 1,000

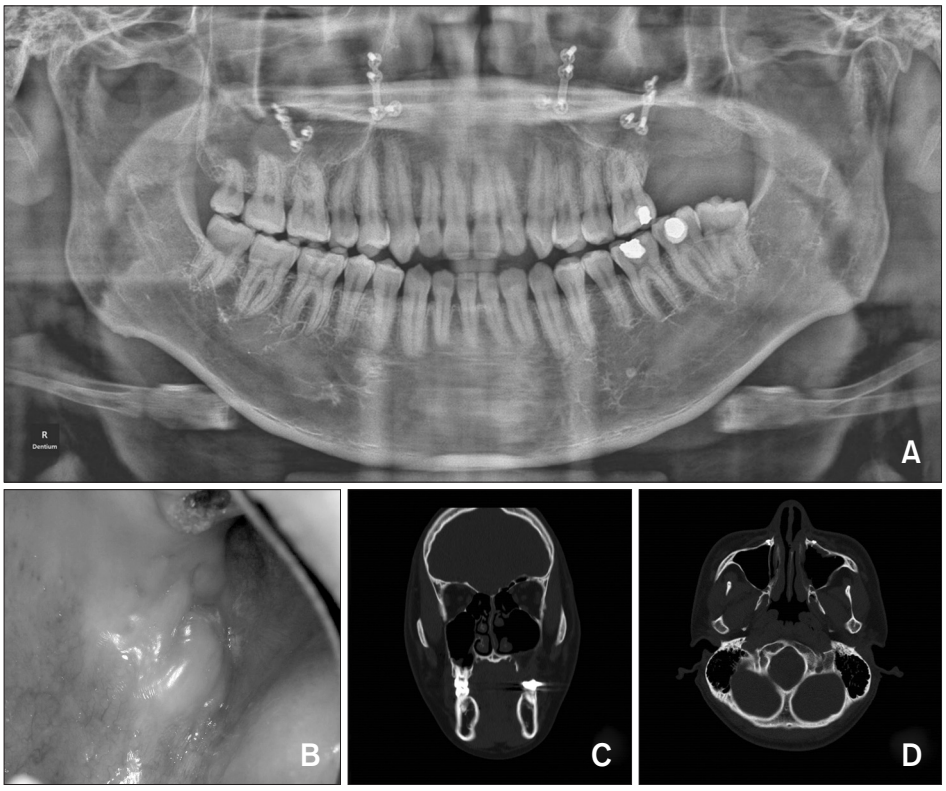


**Fig. 4.** (A) Follow-up panoramic radiograph obtained 3 months postoperatively. No bony healing was observed but the left sinus haziness appears to have been resolved. (B) Follow-up intraoral photograph obtained 1 month postoperatively. The perforated mucosa covered with the pedicled buccal fat pad shows good healing without a fistula. (C, D) Follow-up computed tomography scans in the coronal and axial views obtained 3 months postoperatively. No signs of recurrence was observed.

patients. The patient was discharged from hospital within a week and showed rapid recovery. Over a 6-month follow-up period, radiological and clinical findings showed no aesthetic appearances, no functional impairments, and no sign of recurrence (Fig. 4). These are advantages of Le Fort I osteotomy approach and this approach could be a good option for the treatment of maxillary ameloblastoma. However, additional follow-up examinations are required to monitor the possibility of recurrence.

In conclusion, the maxillary acanthomatous ameloblastoma is a rare case and radical treatment method could affect patient's quality of life. Although ameloblastomas shows high recurrence rate, conservative surgical treatment could be the first choice considering fast recovery.

## Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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