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## Case Report

# A case of ameloblastic fibro-odontoma occupying the left maxillo sinus which was enucleated by the Le Fort I osteotomy

Saburo Tsuruki<sup>a,\*</sup>, Kaneko Kousuke<sup>a</sup>, Yoshimi Ichinokawa<sup>a</sup>, Takashi Tsuruki<sup>b</sup><sup>a</sup> Department of Plastic, Oral and Maxillofacial Surgery, Teikyo University School of Medicine, 2-11-1 Kaga, Itabashi, Tokyo 173-8606, Japan<sup>b</sup> Tsuruki Clinic Medical Dental, 1-19-13 Yawata, Ichikawa, Chiba 272-0021, Japan

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## ABSTRACT

Ameloblastic fibro-odontoma resembles ameloblastic fibroma but it differs in that it contains dentin and enamel. This is a very rare type of odontogenic tumor that occurs most commonly in the posterior mandible. We report a case of 31-year-old female in whom an extremely large ameloblastic fibro-odontoma appeared in the left maxilla and extended to the orbital floor. We removed the tumor by means of Le Fort I osteotomy. We obtained esthetically good results without recurrence.

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## 1. Introduction

Le Fort I osteotomy is the conventional method now widely applied for orthognathic surgery, although it was originally applied for enucleating the tumor in maxilla by Von Langenbeck in 1861 [1]. We treated a 31-year-old female patient who had ameloblastic fibro-odontoma (AFO) in the left maxilla, extending to the orbital floor. We removed AFO by Le Fort I osteotomy and we obtained good results. This led us to believe that Le Fort I osteotomy has several advantages in removing the tumor. We explain how we arrived at this conclusion and supply with biographical information.

## 2. Case report

The patient was a 31-year-old female. The patient had an initial discomfort in the upper left jaw at the beginning of 2008 and noticed some swelling from time to time but ignored the symptom. In August 2010, the patient had the left upper 2nd molar extracted at a nearby clinic. A histo-pathologic examination of

the extracted left upper 2nd molar and its surrounding soft tissue showed ameloblastoma. A second opinion was obtained at another clinic. The latter clinic referred the patient to our hospital (Teikyo University Hospital).

No specific illness was found after a full physical examination. We noted that the left side of the face appeared slightly swollen. There was no squint and the patient did not suffer from double vision (Fig. 1A and B). Swelling on the left upper alveolus was noted. We found a tumor in the socket made as a result of the extraction of upper left 2nd molar (Fig. 1C). X-ray and CT showed the presence of an impacted tooth on the orbital floor. Root resorption of the upper left first molar was recognized by panoramic X-ray (Fig. 2A). CT examination indicated a unilocular neoplastic lesion with swelling of the left maxilla. This lesion extended from the floor of the left maxillo sinus to the orbital floor. We were able to identify the presence of an impacted tooth very near to the orbital floor (Fig. 2B). The posterior of the maxilla was swollen due to the growth of the lesion. We also found the presence of formations of fragments of hard tissues around the impacted tooth (Fig. 2C). The case was clinically diagnosed as ameloblastoma of maxilla. After the extraction of the upper 2nd molar for histopathological examination at the first clinic, the socket was left open. In December 2010, we extracted the tumor under general anesthesia. The incision was made into the oral vestibule which is the general practice for the Le Fort I osteotomy. Periosteum excision revealed that there was no loss of front wall of the maxilla. As a conventional method, osteotomy of the maxilla was performed from the piriform aperture. Prior to the Le Fort I fracture, a spatula was inserted to avoid the damage

<sup>\*</sup> Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

\* Corresponding author. Tel.: +81 047 334 0030; fax: +81 047 334 0029.

E-mail addresses: [wflipb@yahoo.co.jp](mailto:wflipb@yahoo.co.jp) (S. Tsuruki),

[kousuke08232000@yahoo.co.jp](mailto:kousuke08232000@yahoo.co.jp) (K. Kousuke), [yoshimi@med.teikyo-u.ac.jp](mailto:yoshimi@med.teikyo-u.ac.jp) (Y. Ichinokawa), [clinic@tsuruki.org](mailto:clinic@tsuruki.org) (T. Tsuruki).



**Fig. 1.** Initial examination (set of photos). (A) Front view of face. (B) Axial ligation of face. (C) Inside oral cavity. Tumor extruding from left upper 2nd molar socket.

to the eyeball. Using the maxilla separator, we conducted the Le Fort I fracture (Fig. 3A). We were able to excise the tumor from the bone surface of the maxilla using the periosteum raspator. Though the part of the tumor was adhered to the root of the left upper first molar, we were able to remove the tumor as one mass (Fig. 3B). Following this, we extracted the left upper 1st molar, and then removed the remainder of the tumor that had extruded from the socket after the extraction of the left second upper molar. When removing the tumor, we found no sign of bone deficiency in the posterior wall of the maxillo sinus and the orbital floor. After removing the tumor, we performed cryosurgery using liquid nitrogen on the bone surface and soft tissues surrounding the extracted area. Following cryosurgery, we reimplanted the resected bone fragments in the original position and performed osteosynthesis of the maxilla using a titanium plate and titanium screws (Fig. 3C).

Post-operative regular checks were conducted using CT and MRI, and so far the patient is recovering well and there is no sign of recurrence (Fig. 4).

The specimen, measured 48 mm × 48 mm × 29 mm, was nearly oval in shape. The surface texture was smooth and covered with one layer of membrane (Fig. 5).

The odontogenic epithelium was identified as growing in a funicular, sheet-like, net-like way, presenting both anastomotic

and clustering tendencies. At the edge of the epithelial nest the core appears in a palisade arrangement, and inside the alveolar, there are many spaces between the few cells which are star-shaped. Stromal tissue which is fibrous or mucoid and sparse was determined to be a component of the tumor. A part of the epithelial nest is differentiated from the gingiva, as well as from the hard tissues such as dentin enamel (Fig. 6A and B). Histopathologically, the case was diagnosed as ameloblastic fibro-odontoma.

### 3. Discussion

Ameloblastic fibro-odontoma has histologic features of ameloblastic fibroma in conjunction with the presence of dentin and enamel. Clinically, it is asymptomatic and may be detected as a result of a failure of tooth eruption. Ameloblastic fibro-odontoma is often associated with an unerupted tooth [2]. The histologic frequency of occurrence of this type of odontogenic tumor is very low, 0.47%, which makes it a very rare lesion [3]. We were able to confirm that the total number of reports of occurrence of this disease in Japan during the 10 years from 2000 was 18 [4–18]. Thirteen cases were of the lower jaw posterior, one case of lower jaw anterior extending to posterior, one case of upper jaw anterior, and three cases of upper jaw posterior (Table 1).

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