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#### Case report

# Bilateral ramus mandibulectomy with plate reconstruction in ameloblastic carcinoma patient

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# ABSTRACT

**Background:** Ameloblastic carcinoma is a rare and malignant odontogenic tumour possibly arising de-novo from pre-existing ameloblastoma. It is aggressive and locally destructive. Ameloblastoma is the most common benign odontogenic tumour of the mandible. It originates from the tooth-forming epithelium, where its aetiology remains unknown. Ameloblastoma usually grows slowly, is asymptomatic, and destroys the surrounding bone tissue. Malignant transformation of ameloblastomas may occur spontaneously. Resection is the primary therapy for ameloblastic carcinoma with extensive bone destruction. Mandibular resection causes instability due to the missing parts of bone, so reconstruction is needed. **Purpose:** This study will report on an individual case of ameloblastic carcinoma that underwent a bilateral ramus mandibulectomy with reconstruction using the plate technique. **Case:** Bilateral ramus mandibulectomy with plate and reconstruction in an ameloblastic carcinoma patient. **Case Management:** Two months after surgery, the patient could open her mouth functionally and aesthetically. **Conclusion:** Plate reconstruction is an option for reconstructing bilateral ramus mandibulectomy of a large ameloblastic carcinoma of the mandible.

Keywords: ameloblastic carcinoma; ameloblastoma; bilateral ramus mandibulectomy; plate reconstruction

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# **INTRODUCTION**

Mandibular ameloblastoma is a benign odontogenic tumour that is slow-growing and invasive of surrounding structures.<sup>1</sup> This tumour occurs between the ages of 30–60 and has a matching rate of occurrence in males and females.<sup>2,3</sup> Ameloblastoma is divided into unicystic, multicystic, peripheral, and desmoplastic ameloblastoma.<sup>3</sup> The ameloblastoma keeps developing into a malignancy divided into metastatic ameloblastoma and ameloblastic carcinoma.<sup>4</sup>

Ameloblastic carcinoma is a rare and malignant odontogenic tumour that contributes to 1% of all jaw tumours and possibly arises de-novo from pre-existing ameloblastoma.<sup>5</sup> The diagnosis is made by panoramic radiography, computed tomography (CT) scan, and histopathology examination.<sup>6</sup> Management of mandibular ameloblastic carcinoma is by surgery, either conservative or radical. Conservative surgeries include enucleation, curettage, excision, and marsupialization. Radical surgeries include resections such as marginal, segmental, and total resection of the jaw (maxilla/mandible) with wide margins.<sup>7</sup> There is relatively high recurrence with conservative surgery at 60%, whereas radical surgery is at 10%.<sup>1,4</sup> The best chance of healing requires a wide resection with a margin of about 1–1.5 cm.<sup>4</sup>

Post-resection reconstruction aims to restore shape for muscle attachment, chewing, swallowing, speaking functions, and cosmetics. Several techniques commonly used to reconstruct the mandible are osteocutaneous vascularised bone graft, nonvascularised bone grafts, and plate reconstruction with/without soft tissue pedicle flaps.<sup>1</sup> The purpose of this paper is to report on a case of ameloblastic carcinoma that underwent resection and reconstruction using a plate and screw.

# CASE

A 58-year-old woman came into the outpatient clinic with a five-year history of a gradually expanding painless mass in the lower left jaw. The patient could only eat soft food. Physical examination revealed a solitary mass in the left mandibular region measuring 15 cm x 12 cm x 10 cm, causing a marked facial asymmetry. The mass was cystic to touch, fixed, hard consistency, and had normal mucosal colour resembling surrounding tissue. Intraoral examination revealed a solitary mass in the left mandibular region, an almost entirely missing tooth, and no trismus. There were no palpable lymph nodes in the neck (Figure 1).

### **CASE MANAGEMENT**

The panoramic photo showed a primary bone tumour suspected to be chondroblastoma. The CT scan of the head and neck showed a thin septate expansive cystic lesion and a geographic type of destruction. There was a narrow transitional zone, popcorn calcification, and soap bubble appearance of 19 Hounsfield units (HU). Also shown was its size  $\pm 10 \text{ cm x } 8.6 \text{ cm x } 10.1 \text{ cm in the right}$  and left mandibular up to the left mandibular angle with no significant contrast enhancement (23 HU) in the solid

lesion, septa, and floating teeth. The CT scan suggested ameloblastoma or an odontogenic cyst (Figure 2).

The fine-needle aspiration biopsy (FNAB) results revealed a benign cystic lesion. An open biopsy was not performed because the patient refused. The patient was diagnosed with mandibular ameloblastoma, and surgery was scheduled for a bilateral ramus mandibulectomy and mandibular plate reconstruction.

The mandibular resection was carried out on March 14, 2019, and a pre-operative tracheotomy was performed. An incision was made to divide the lower lip from the midline along the mental region backward to the submandibular region. Then a flap was made upwards past the lower border of the mandible to preserve the mandibular ramus and facial nerve. Blunt dissection separated the masseter muscle from the bone and tumour mass. The right and left mandibles were cut with a Gigli saw about 1.5 cm beyond the tumour margin, then the left and right mandibular ramus were left ( $\pm$  2.5 cm). After removing the tumour, cauterization was performed in the centre of the residual bone segment to prevent a recurrence.

Mandibular reconstruction, along with plate and screw, was made of titanium. The plate was shaped according to the mandibular arch and later placed in order occlusion. It was installed with screws on the left mandibular with a two-piece segment and a three-piece segment on the right



Figure 1. Clinical appearance; A) Front view of the face; B) Side view; C) Sublingual mass and the remaining teeth are shown.



Figure 2. CT scan of multicystic head and neck. A) Coronal section; shows the popcorn calcification. B) Sagittal cut shows the soap bubble appearance. C) Axial cut.

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Figure 3. Surgery Stages: (A). Disinfection of the surgical area after tracheostomy and intubation; (B). Separation of tumour mass from nearby organs; (C). The mandible was cut using a Gigli saw; (D). The left mandible resected; (E). The type and size of the plate used (arrow); (F). Inserted plates and screws on the mandible, the left side with three screws and the right side with two screws; (G). Gingivobuccal mucosa suturing; (H). Multicystic tumour weighing 800 grams.



Figure 4. A. 5th postoperative day: (1). Shut mouth; (2). Mouth open about 2 cm wide. B. Two months postoperatively: (1). Shut mouth; (2). Open mouth, max-width.

mandibular. The mylohyoid muscle and soft tissue were sutured around the plate. The gingivobuccal mucosa was sutured to the floor of the mouth using the watertight and airtight principles (Figure 3). Vacuum drains were placed on the right and the left of the submandibular. The operation results showed the mandibular tumour was multicystic, measuring 17 cm x 12 cm x 8 cm, and weighed about 800 grams. Postoperative histopathology was ameloblastic carcinoma with tumour-free resection margins.

Furthermore, the recovery treatment took ten days with nutrition provided through a nasogastric tube. By the fifth day, the surgical wound was dry, and the patient could open her mouth about 2 cm. At two months postoperative, her mouth could be opened normally, and she had oral nutrition intake (Figure 4). We consulted with the patient at the dental and oral clinic, but a denture (prosthetic) could not be installed because there were no teeth for fixation. The patient did not receive chemoradiation because the plate and screw were attached. Fortunately, no lymph node enlargement and tumour-free resection edges were found in the histopathological results. The patient was scheduled for an x-ray evaluation three months postoperatively, but the patient did not appear for control, and contact was lost.

#### DISCUSSION

These tumours could occur in anyone between 30–60 years old. The ratio of occurrence is the same for males and females.<sup>2</sup> The signs and symptoms of mandibular ameloblastoma are swelling of the mandible, painlessness, loose teeth, and even loss of chewing and swallowing disorders.<sup>2,4</sup> The patient in this study was a 58-year-old woman. Chief complaints were a large lump in the lower left jaw that was painless and incomplete teeth. Based on FNAB results, benign cystic lesions were shown. Furthermore, the CT scan showed ameloblastoma and odontogenic cysts. The CT scan of a multilocular type of ameloblastoma showed a classic soap bubble appearance.<sup>8</sup> The patient was diagnosed with mandibular ameloblastoma.

In case reports of the bilateral ramus, mandibulectomy is performed from the left mandibular ramus to the right. A bilateral bounded mandibulectomy is a resection of the anterior aspect of the mandible crossing the midline with an intact posterior mandibular segment bilaterally. It is further divided into five classes descriptively, one of which was bilateral ramus mandibulectomy.<sup>9</sup> In this case, resection was performed about 1.5 cm outside the tumour with the remaining bone segments, with the right and left mandibular ramus  $(\pm 2.5 \text{ cm})$  as the plate placement. After the tumour was removed, the centre of the residual bone segment was cauterized. The best treatment option for ameloblastoma was a radical excision of the tumour mass, reaching the normal bone with a tumour-free margin of 1-2 cm.<sup>10,11</sup> To prevent a recurrence, procedures were added by adjuvant therapeutic. This therapy was performed on the intraoperative bone margin through tissue fixation, drilling, or cauterizing bone tissue.<sup>8</sup>

There was also mandibular reconstruction with a plate and screw in this case. A fibula flap is considered the gold standard of choice owing to its length, bone stock, reliable pedicle, tolerance of dental implants, and low donor-site morbidity. Although the free fibula flap provides rigid support, a soft tissue flap with a bridging plate, on the other hand, is often easier to perform as it allows for sample soft tissue and easily achieves defect closure.<sup>12</sup> Mandibular reconstruction was done using a plate and screw made of titanium and shaped to match the patient's mandibular arch. The screws inserted were two pieces on the left and three on the right of the mandibular segment. Mandibular reconstruction plates and screws are the most widely used alloplastic devices for mandibular reconstruction. The most common metals fabricating these plates are stainless steel, vitallium, and titanium.13

In the reconstruction stage, the operator formed a plate with a hand-forming technique that matched the contour of the patient's bone. The ideal reconstruction would provide a solid arch to articulate the upper jaw that would restore swallowing, speech, mastication, and aesthetics.<sup>11</sup> The mylohyoid muscle and soft tissue were sutured around the plate. The gingivobuccal mucosa was sutured to the mouth floor with watertight and airtight principles. Later, the technique of suturing muscle and soft tissue, fixation of internal soft tissue, mucosa, and suturing the skin surface properly and correctly will affect the success of the function of the masticatory muscles in postoperative patients..<sup>14</sup>

In this case, there were differences in the pre and postoperative histopathological results. Preoperatively, the ameloblastoma was determined. Consequently, treatment was planned for a benign tumour. At the same time, the postoperative histology results showed ameloblastic carcinoma with tumour-free resection edges. Ameloblastic carcinoma represents a malignant transformation of pre-existing well-differentiated ameloblastoma or odontogenic cyst.<sup>15</sup> Because of the location of the plate and screw reconstruction, chemoradiation was not recommended. Radiotherapy can be suggested to decrease tumour size before surgery and to improve local control when surgical margins are close or microscopically positive. Experience with chemotherapy is minimal in treating ameloblastoma and is primarily limited to isolated cases.<sup>11</sup>

Later, no enlarged lymph nodes were shown, with postoperative histopathological results of tumour-free resection ends. For patients with local recurrence or inadequate margins after surgery, adjuvant radiotherapy provides the potential for disease control.<sup>16</sup> The patient was recommended to have regular control to detect recurrence and distant metastases. If recurrence and metastases occur, resection will be carried out.<sup>13</sup> This technique was simple and uncomplicated and had a satisfactory success rate. This technique's success rate is high without requiring microscopes, special techniques, or microsurgery.

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