A Case of Central Carcinoma of the Mandible Arising from a Recurrent Odontogenic Keratocyst :
Delineation of Surgical Margins and Reconstruction with Bilateral Rectus Abdominis Myocutaneous Free Flaps

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A case of central carcinoma of the mandible arising from a recurrent odontogenic keratocyst is reported. A 38-year-old man was admitted to the Tokai University Hospital due to postoperative infection of a recurrent odontogenic keratocyst of the left mandible. He had had a cystectomy for an odontogenic keratocyst 4 years ago. The lesion revealed bony destruction of the mandible with worm-eating shaped margins with extension to the facial skin. A biopsy specimen revealed squamous cell carcinoma. The mandible was resected with facial skin and the sublingual space was dissected to preserve the lingual nerve. The oral and the facial resections were reconstructed with a titanium plate and bilateral rectus abdominis myocutaneous free flaps. The plate was removed due to infection around the margins and readjustment of the flaps was conducted 5 months after the surgery. He has not had a local relapse, metastasis, or incisional hernia for 8 months following surgery. Good occlusion has been attained by the residual mandible, and he is able to eat without any problems.

Key words : Central carcinoma of mandible, Odontogenic keratocyst, Surgical margins, Rectus abdominis myocutaneous flap
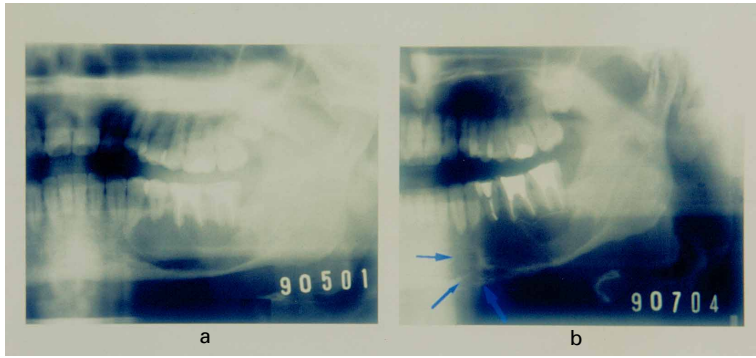
INTRODUCTION

Central carcinoma of the mandible is an uncommon lesion. This tumor may theoretically arise 1) from the lining of odontogenic cysts, 2) from other epithelial odontogenic tumors, or 3) de novo from presumed odontogenic rests. We experienced a case of central carcinoma of the mandible arising from a recurrent odontogenic keratocyst that invaded the facial skin. The presentation, delineation of surgical margins, and method of reconstruction are discussed.

CASE REPORT

A 38-year-old man was admitted to the Tokai University Hospital due to postoperative infection of a recurrent odontogenic keratocyst of the left mandible on September 11, 1997. He had undergone a cystectomy in June 1993 for an odontogenic keratocyst.

After the operation, he was not followed up. In April 1997, he felt a swelling in the mandible and visited the oral surgeon who had operated on him. Orthopanoramic tomography showed an oval radiolucent area in the left mandible, the margins were smooth in shape, and the apex of the first and second premolars were not absorbed. (Fig. 1a). He was operated on again on May 22, 1997 for a recurrent odontogenic keratocyst. He complained of hypesthesia of his lower lip on the left side after the operation. The surgical site became infected seven days after the operation and was irrigated daily. However, despite treatment, the infection continued and a fistula was found on June 5. Orthopanoramic tomography revealed marked destruction of bone tissue with worm-eating shaped margins (Fig. 1b). On admission, there were no abnormal intraoral findings and the fistula found in the sub-

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Fig. 1 Orthopanoramic tomography of the mandible. **a** ; May 1, 1997, the margins are clearly defined and the apex of the premolars are not absorbed. **b** ; July 4, 1997, note the marked destruction of bone tissue with worm-eating shaped margins.

Fig. 2 Intraoral and facial findings.
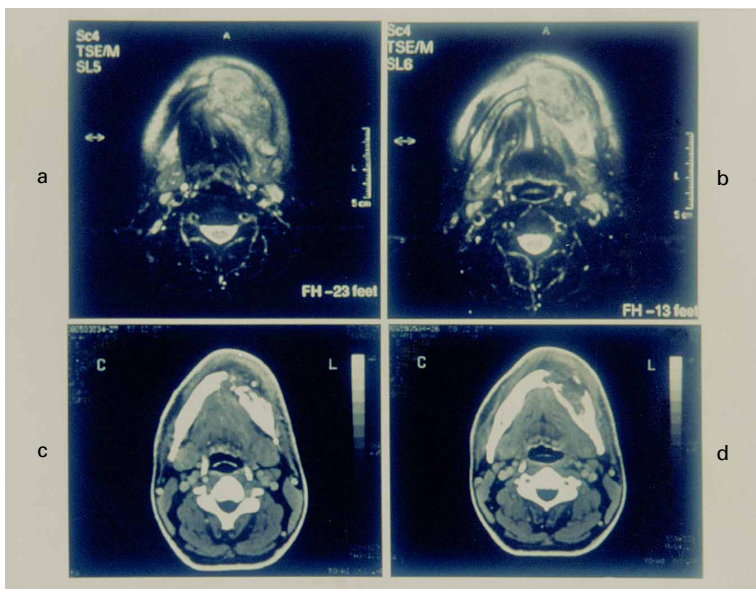
Fig. 3 a, b MRI shows a mass lesion that extends from the skin of the cheek to the sublingual space (high signal intensity T2 image).
      c, d CT shows destruction of mandibular cortical bone.
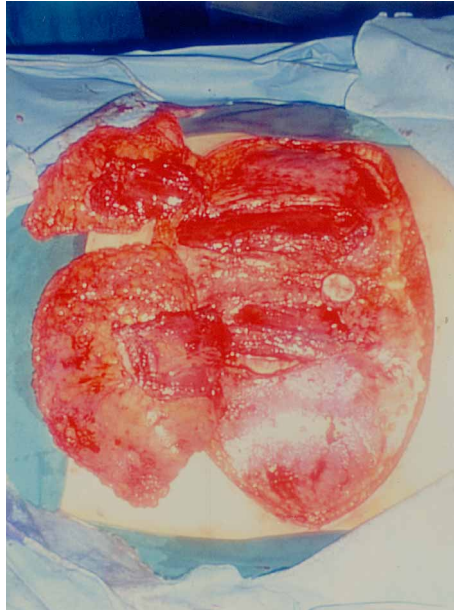
Fig. 4 Bilateral RAMC flaps.

mental region was associated with cutaneous redness (Fig. 2a, b). Although ampicillin was administrated to treat the infection, no improvement was noted. Computed tomography (CT) showed destruction of the buccal and lingual side of the mandibular cortical bone (Fig. 3c, d). Magnet resonance imaging (MRI) showed a lesion that extended from the skin of the cheek to the sublingual space on the high signal intensity T2 image (Fig. 3a, b). The first and second premolars were extracted and tissue from the dental alveoli was histopathologically examined for the presence of malignant disease, but a diagnosis of malignancy was not confirmed. However, a biopsy from the submental region of the fistula revealed squamous cell carcinoma. The patient was operated on under general anesthesia on July 30. The surgical margins on the cheek and submental skin were delineated one centimeter outside the area invaded by the carcinoma, as determined by ultrasonography. The mandible was resected from the right second premolar region to the left ramus and the sublingual space was dissected to preserve the lingual nerve.

The primary lesions and excised cervical lymph nodes were resected as one lump.

A titanium plate was used in the mandibular reconstruction. Bilateral rectus abdominis myocutaneous flaps (RAMC flaps), fed by the inferior epigastric artery, were grafted to the oral and facial sites of resection. In conducting microangioanastomosis, the facial artery and the facial vein were used for oral reconstruction and the superior thyroid artery and the superior thyroid vein were used for facial reconstruction (Figs. 4, 5, 6). The initial closure of the donor site of the RAMC flaps was conducted. The operation lasted nine hours and fifty minutes; estimated blood loss was 417 ml.

Although the patient had an uneventful postoperative progress for 5 months after the surgery, the area around the reconstruction plate became infected. The plate was removed and debriedment was conducted. The bulk of the flaps was also readjusted. He has not had a local relapse, metastasis, or incisional hernia for 8 months following surgery. Good occlusion has been attained by the residual mandible and he eats satisfactory.

Histologic Description

In the surgical specimen obtained at the first operation on June 1993, the cyst wall showed a lining of keratinized stratified squamous cells without rete ridges and a thin fibrous tissue with mild inflammatory cell infiltration. The luminal surface of the
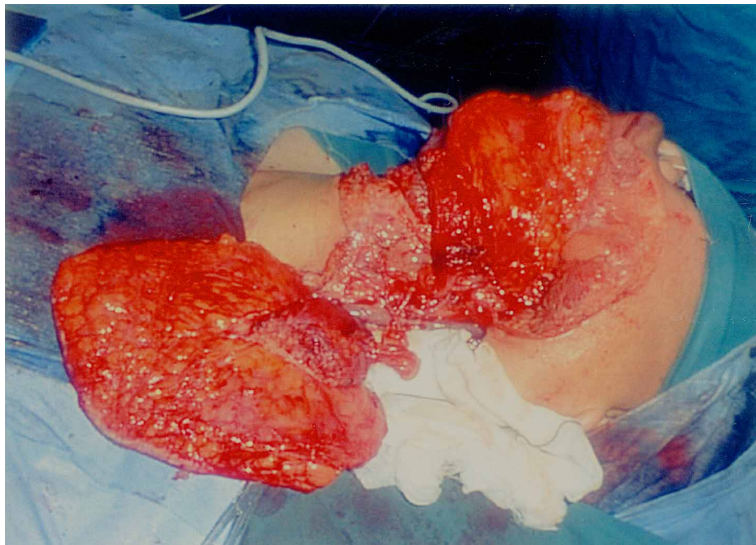
Fig. 5 After microangioanastomosis and reconstruction of the oral site.

**Fig. 6** After reconstruction of the face.

keratinized epithelium was corrugated and hyperkeratosis predominated over parakeratosis. Uneven thickening of the spinous layer was seen. The spinous layer region had a prominent granular layer. There were neither islands of epithelium nor separate daughter cysts in the fibrous tissue of the cyst wall. Based on these findings, the cyst was diagnosed as an orthokeratinized variant of the odontogenic keratocyst (Fig. 7).

The surgical specimen obtained at the sec-ond operation revealed comparable findings.

The third biopsy specimen showed invasive growth of atypical squamous cells with keratinization and epithelial pearl formation, and thus the lesion was diagnosed as a well differentiated squamous cell carcinoma.

The extensively resected mandibular bone showed an invasive, bony destructive lesion, approximately 2 cm in diameter (Fig. 8). The specimens dissected from the main tumor showed a mass of squamous cell carcinoma
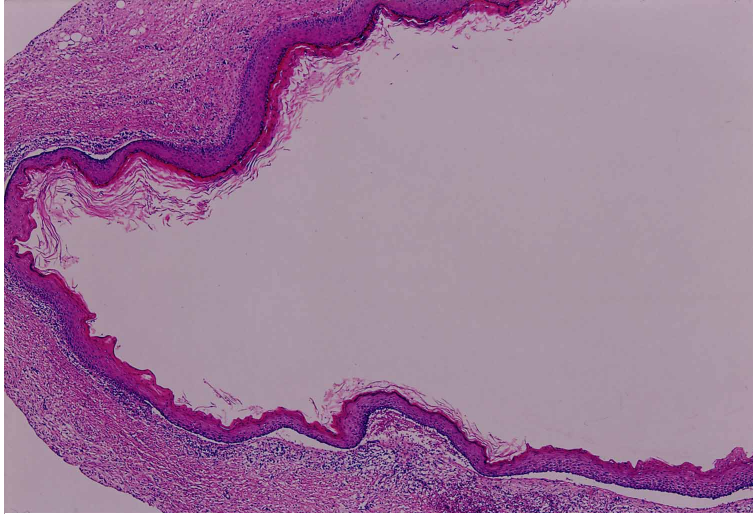
Fig. 7 Low power view of the thin orthokeratinized epithelium of the cyst wall and the corrugated appearance of the inner cystic surface. No epithelial islands or daughter cysts are seen in the cyst wall (HE, X10)
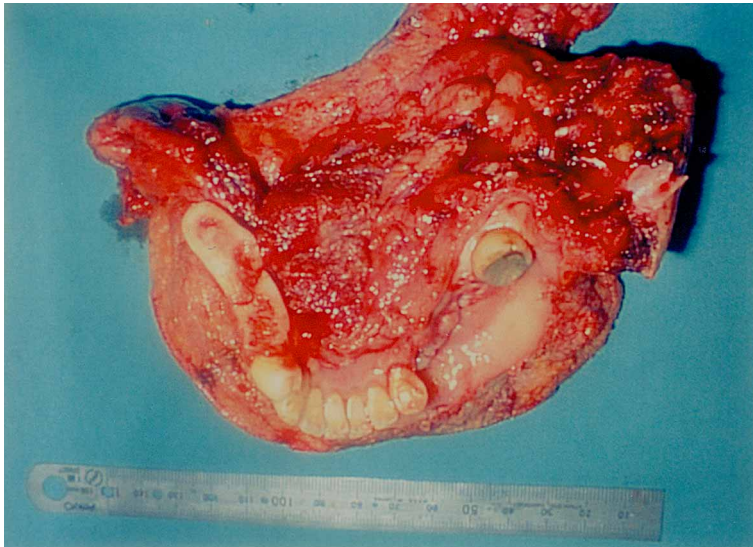

Fig. 8 Macroscopical findings of the tumor.

with marked keratinization and cancer pearl forms. Part of the tumor was arranged in an odontogenic pattern, with basal-type cells forming alveoli. The tumor had completely destroyed the cortical bone on the buccal and lingual side and invaded the surrounding fibrous tissue, but apparently it did not invade the muscles and sublingual glands. Metastatic carcinoma was not seen in any cervical lymph node. A cyst wall, associated with the orthokeratinization type of odonto-genic keratocyst, was revealed in the tumor. The luminal surface of the epithelium showed marked keratinization. A transitional region was identified between the cyst wall and the carcinoma. These histological features and history of recurrent odontogenic keratocyst led us to diagnose this lesion as a squamous cell carcinoma arising from a malignant change in the odontogenic keratocyst (Figs. 9, 10).
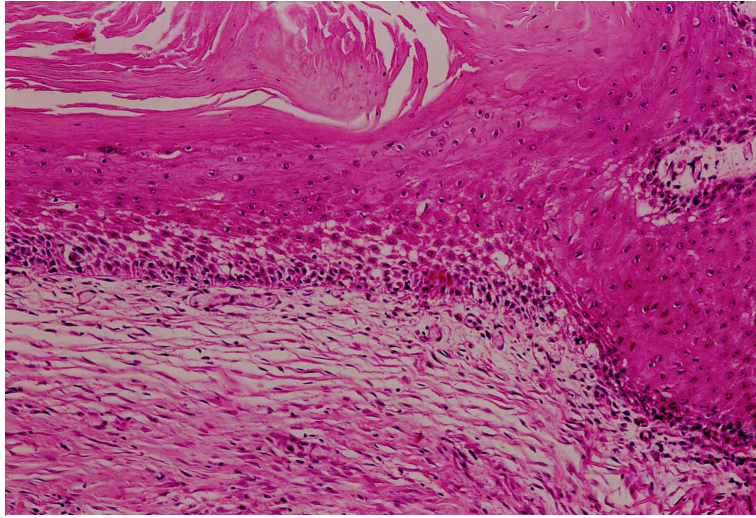
Fig. 9 Lower power view showing the transition of the cyst wall into carcinoma. The invasion of neoplastic transformed epithelium into the submucosal stroma is visible.
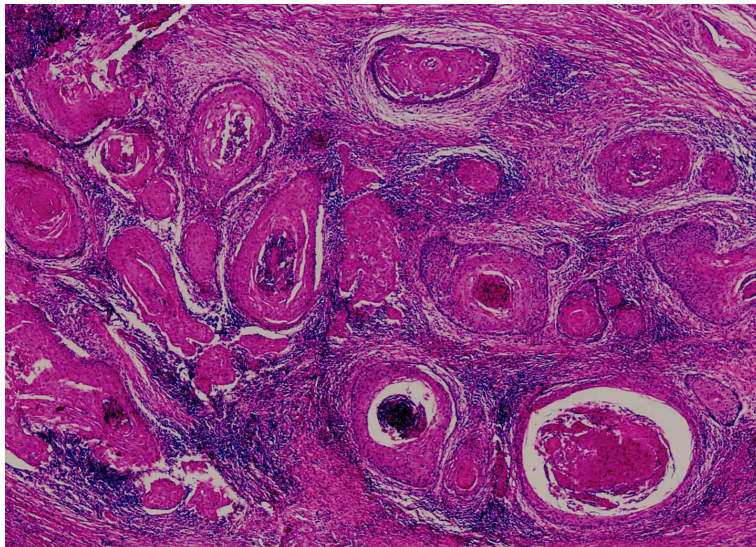

Fig. 10 Lower power view shows proliferation of atypical squamous cells, which are frequently keratinized, and cancer pearl formation.

DISCUSSION

[Pathology]

1. Odontogenic carcinomas may develop by malignant transformation of an ameloblastoma, directly from residues of odontogenic epithelium following tooth development, or from the epithelial lining of odontogenic cysts [1, 2]. In the above, odontogenic carcinomas developing from the epithelial lining of odontogenic cysts are histologically classified as squamous cell carcinomas. We were able to find only one reference in the literature reporting a carcinoma arising from a recurrent odontogenic cyst [2]. Therefore, it is necessary to clinically demonstrate that there is no connection between tumor and oral mucosa, and to rule out the possibility of metastasis from another site [3]. Furthermore, if the transforma-
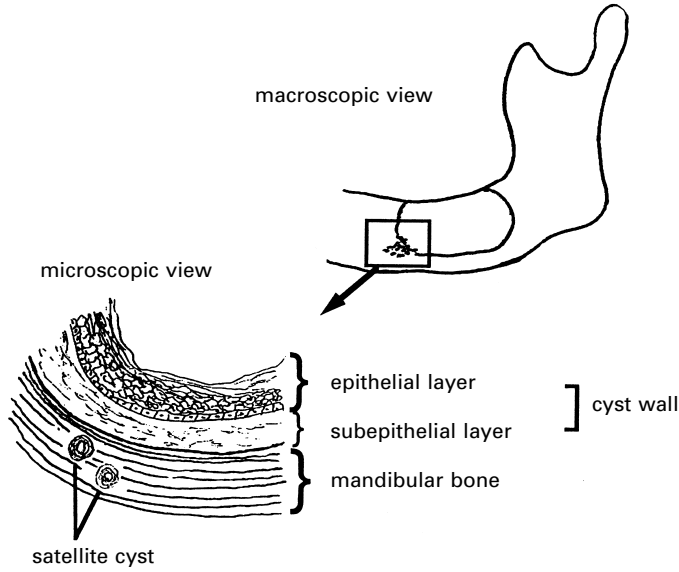
Fig. 11 Schematic view of satellite cysts.
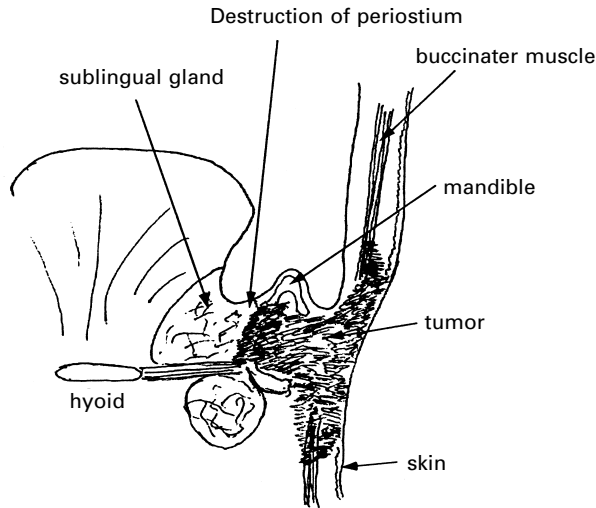
Fig. 12 Schematic view of the area invaded by carcinoma cells.

tion from cystic epithelium to squamous cell carcinoma can be verified, it is possible to make the diagnosis of squamous cell carcinoma originating from an odontogenic cyst. In our case, the patient twice suffered a relapse of an odontogenic keratocyst, and the transformation from odontogenic keratocyst to squamous cell carcinoma was seen. Odontogenic keratocysts are classified into two types, parakeratosis and hyperkeratosis, based on the type of keratinization. Of these two types, the parakeratotic odontogenic keratocyst is highly proliferative. Satellite cysts or islands of odontogenic epithelium are observed in connective tissues in many cases, indicating that the cyst is highly likely to relapse. In our patients, we thought that malignant transformed satellite cysts were already in the mandibular bone at his second cystectomy (Fig. 11). The patient had a

hyperkeratotic odontogenic keratocyst which is generally accepted to be less proliferative and therefore less likely to relapse [4]. However, the patient had two relapses necessary to monitor progress even if the odontogenic keratocyst is histologically classified as hyperkeratotic.

[Delineation of Surgical Margin]

Invasion into surrounding soft tissues and the destruction of the buccal and lingual sites of the mandibular cortical bone was seen in this case. Very little exists in the literature because central carcinoma of the mandible is a rare disease. To delineate the surgical margin, standards for carcinoma of the lower gingiva invading the mandible and surrounding soft tissues were applied to this case, based on the patient's well differentiated squamous cell carcinoma and finding that invasion into the surrounding soft tissues had occurred. As for treating the bone in cases of carcinoma of the lower gingiva, there are numerous reports delineating the surgical margin according to the depth and form of bone absorption [5]. However, no criteria for delineating the surgical margin of soft tissues are not available. Kishi *et al.* [6], conducted a study on prognosis, based on the classification of carcinoma of the lower gingiva into two types; mucosa of the oral floor and the type of buccal mucosa, according to the form of invasion into soft tissues. They suggested that stump recidivation recurrence was more likely to occur in soft tissues than hard tissues (mandible) and emphasized the importance of treatment of the soft tissues [6]. However, submucosal invasion is seen in many cases of oral cancer even though they are clinically classified as early cancer [7]. Therefore, in identifying the progress of carcinoma of the lower gingiva that has extended the buccal mucosa or the mucosa of floor of the mouth, not mucosal invasion but three-dimensional invasion should be observed. As for treatment of the bone, because the patient had central carcinoma of the mandible and that bone absorption was seen as far as the inferior mandibular border, segmental resection was performed. The surgical margin was delineated two centimeters beyond the border of the spread of bone absorption shown by orthopanoramic tomography. As for the treatment of the soft tissues, since invasion was into the face and skin in the mental region on the buccal side, the surgical margin of the skin was delineated by means of ultrasonography. It has been suggested that ultrasonography and histopathology reveal similar borders of invasion in many cases of oral cancer [8]. In our patient, the surgical margin of the skin was delineated one centimeter outside the invasive lesion, as shown by ultrasonography. No stump tumor invasion was seen by histopathology. Absorption in the lingual side of the mandibular cortical bone was seen, and the tumor was connected with the sublingual space. In the T2 intensified MRI scan, the whole sublingual space was noted as a high signal intensity area. The observation which indicated invasion was seen in the sublingual space in CT. The area invaded by the carcinoma is shown in Figure 12. Based on these findings, dissection of the sublingual space was conducted with the aim of preserving the lingual nerve. In the dissection of the sublingual space, the mucosa of oral floor was concurrently resected because of nonexistence of true fascia between the sublingual gland and the mucosa of the oral floor [9]. In the histological sections, carcinoma cells were not seen in the sublingual gland. However, it was decided to resect the mandible with dissection of the sublingual space, because we believed that the carcinoma cells that destroyed the periostium of the mandible could easily disseminate in the sublingual space. Furthermore, because absorption in the lingual side of the mandibular cortical bone extended to the inferior border of the mandible and indicated dissemination to the submandibular space, it was considered important to resect the primary focus and excise the cervical lymph node simultaneously. This was thought to be optimal for achieving a complete curve. The patient has satisfactory buccal function as a result of conservation of the lingual nerve and reconstruction of the oral floor.

[Method of Reconstruction]

In the reconstruction of the oral mucosa and facial skin, which required two islands of skin, we proceeded as follows: (1) to secure the islands of skin by de-epithelization of part of either the unilateral rectus abdominis myocutaneous or latissimus dorsi myocutaneous (LDMC), (2) to use the LDMC

and scapular flaps fed by the same thoracodorsal artery or (3) to use completely different flaps such as the fore arm and RAMC flaps. In the case of (1), the facial and oral reconstruction requires bending the flaps, which can cause partial necrosis resulting from impaired blood supply to the periphery of the flaps. The adjustment of bulk is also a problem at this time. In the case of (2), the LDMC and scapular flaps are usually fed by the same vessel, but in some cases by different vessels, a situation which is incompatible with this surgical technique under which microangioanastomosis is conducted at one site and two flaps are supplied. The surgery of oral cancer is usually conducted with the patient in the supine position. This means that reconstruction using flaps from the dorsal region requires two postural changes, which prolongs the surgical time by at least one hour because of difficulty in securing an aseptic surgical field. It is difficult to conduct initial closure of the surgical wound if a wide area of flaps is collected from the dorsal area. This, therefore, usually requires a split thickness skin graft, and the patient will have yet one more surgical wound. Furthermore, we instruct patients who have undergone microangioanastomosis to rest in bed for 3 days after the procedure, and the existence of a surgical wound in the dorsal area increases their pain. Procedure (3) entails increased surgical wounds as well as microangioanastomosis at two sites.

For our patient, we conducted oral and facial reconstruction using bilateral RAMC flaps. Bilateral RAMC flaps are commonly used as pedicle flaps for reconstruction after mastectomy [10]. However, there are no reports concerning reconstruction of the head and neck region using such flaps as free flaps. With this technique, it is relatively easy to adjust of bulk since each side of the flap is grafted separately. It is also possible to provide an initial closure at the donor site. Because our patient had little postoperative contraction of the flap used as facial skin, and had an infection arising from the titanium plate, additional surgery was necessary to remove the plate and adjust the bulk 5 months after the first operation. Although it is suggested that patients are likely to develop incisional hernia as a postoperative complication [10], our patient was no longer bed-ridden by Day 5 after surgery and was

free of any other abnormality. With this technique, although the patient will have surgical wounds at different sites of the head, neck, and abdomen, excision of the tumor and collection of RAMC flaps can proceed concurrently once the resection volume in the head and neck is decided. The concurrent closure of donor sites and microangioanastomosis and reconstruction in the head and neck also contributes to a reduction in surgical time. Taking advantage of this time-saving procedure, it was possible to reconstruct the mandible, oral mucosa and facial skin in 9 hours and 15 minutes. Though the technique requires microangioanastomosis at two sites, we have reached the conclusion that the technique is useful in reconstruction of the head and neck.

REFERENCES

1) Waldron C. A. and Mustoe T. A. : Primary intraosseous carcinoma of the mandible with probable origin in an odontogenic cyst. Oral Surg. Oral Med. Oral Pathol. 67 : 716-724, 1989.
2) Dabbs D. J., Schweitzer R. J., Schweitzer L. E., and Mantz F. : Squamous cell carcinoma arising in recurrent odontogenic keratocyst : case report and literature review. Head & Neck 16 : 375-378, 1994.
3) Pindborg J. J., Kramer I. R. H., and Shear M. : Histological typing of odontogenic tumors. In : 2nd ed. WHO international histological classification of tumors. Springer-Verlarg, Berlin, Heidelberg, New York, London, Paris, Barcelona, Budapest. : 24-26, 1992.
4) Regezi J. A., and Sciubba J. J. : Cysts of the oral region. In : Oral pathology. W. B. Saunders Co.. Philadelphia. 313-317, 1989.
5) Kusama M., Kishi T., Hosho K., et al. : Clinical Study on method of resection for carcinoma of lower gingiva. Head & Neck Cancer 23 : 631-636, 1997.
6) Kishi T., Kusama M., Horikoshi M., et al. : The clinical study on surgical treatment of squamous cell carcinoma of the lower alveous and gingiva. —The extent of primary lesion and cervical metastasis— Jpn. J. Oral Maxillofac. Surg. 37 : 633-641, 1991.
7) Steinhart H., and Kleinsasser O. : Growth and spread of squamous cell carcinoma of the floor of the mouth. Eur. Arch. Otorhinolaryngol. 250 : 358-361, 1993.
8) Kato T., Hayashi T., and Munakata R. : Evaluation of ultrasonography with intraoral scanner in T1 and T2 carcinoma of the tongue. Jpn. J. Oral Maxillofac. Surg. 43 : 809-813, 1997.
9) The oral cavity. Emphasizing the sublingual and submandibular spaces. In : Harnsberger H. R. ed. 2nd Handbook of Head and neck imaging, Mosby, New York, Philadelphia, Sydney, Tokyo, Tront 1995.
10) Nishimura M. : Immediate breast reconstruction — Using rectus abdominis myocutaneous flap. Rynshou Geka 47 : 1321-1331, 1992.