Bilobed Flap for Reconstruction of Skin Defects after Excision of Parotid Carcinoma: A Case Report

Koji Sakamoto^{1,2}, Hiroyuki Ozawa³, Masayuki Shimoda⁴, Masashi Nakaishi², Atsuko Sakanushi², Takeshi Matsunobu², Kimihiro Okubo², Takashi Okada¹, Yoichiro Sato¹, Seiichi Shinden¹ and Kaoru Ogawa³

¹Department of Otorhinolaryngology, Saiseikai Utsunomiya Hospital, Tochigi, Japan ²Department of Otorhinolaryngology, Head and Neck Surgery, Nippon Medical School, Tokyo, Japan ³Department of Otorhinolaryngology, Head and Neck Surgery, Keio University, Tokyo, Japan ⁴Department of Pathology, Keio University, Tokyo, Japan

Objectives: Local flaps, pedicled flaps, and free flaps are used to reconstruct medium-sized skin defects after excision of parotid carcinoma. The bilobed flap is a local flap primarily used by plastic surgeons for small defects of nasal skin. We report a case of parotid carcinoma with skin infiltration successfully treated by skin reconstruction with a bilobed flap.

Methods: An 84-year-old man presented with a parotid mass he had noticed 2 months earlier. Parotid carcinoma with skin infiltration was diagnosed and he underwent radical surgery. The skin defect was round (diameter, 6 cm) and was resected and reconstructed with a bilobed flap designed to be caudal to the defect.

Results: Postoperative facial nerve palsy improved within 6 months. The postoperative course was otherwise uneventful, and the patient was discharged on postoperative day 7. Pathological examination revealed a sarcomatoid salivary duct carcinoma.

Conclusions: Bilobed flaps are useful for reconstructing skin defects with a diameter of 6 cm or less. (J Nippon Med Sch 2022; 89: 606–611)

Key words: bilobed flap, parotid gland malignancy, local flap, sarcomatoid salivary duct carcinoma, geriatrics

Introduction

Parotid carcinoma tends to invade surrounding structures, such as subcutaneous tissue, skin, cartilage of the external auditory canal, mandibular bone, and facial nerve. In cases of skin invasion, the involved skin must be excised. However, skin stretched by a slow-growing tumor or small defect is closed by reefing with the remaining skin. When the defect cannot be closed, skin reconstruction is required for rapidly growing, high-grade, malignant parotid tumors with direct skin involvement.

Skin reconstruction methods comprise local flaps, pedicled flaps, and free flaps. Selection of the appropriate flap requires consideration of the site and range of the

defect and the patient's general condition, including age and comorbidities. Studies have reported reconstruction using bilobed flaps for skin defects after tumor resection in the infraorbital, ear, and temporal regions^{1,2}. However, to our knowledge, no study has reported using a bilobed flap for reconstruction after excision of parotid carcinoma. Herein, we report a case of parotid carcinoma with skin infiltration successfully treated by skin reconstruction with a bilobed flap.

Case Report

An 84-year-old man was referred to our hospital with a right parotid mass, which he had noticed 2 months ear-

Correspondence to Koji Sakamoto, Department of Otorhinolaryngology, Head and Neck Surgery, Nippon Medical School, 1–1–5 Sendagi, Bunkyo-ku, Tokyo 113–8603, Japan

E-mail: k-sakamoto@nms.ac.jp

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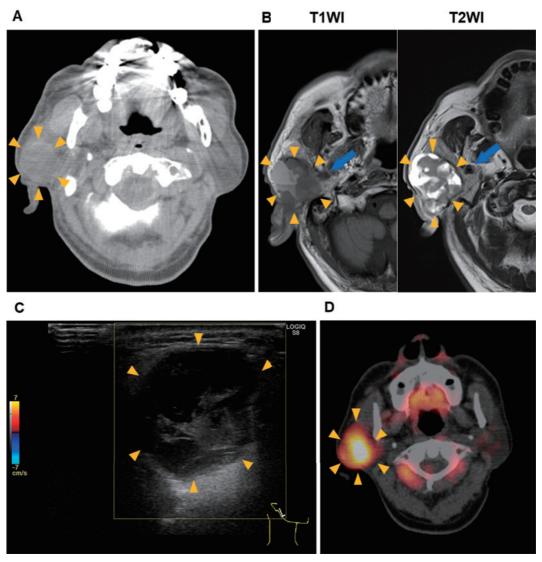


Fig. 1

- A: Axial CT scan showing an almost isodense mass in the right parotid gland.
- B: Axial MRI scans. T1WI scan of the right parotid gland showing a hypointense to isointense mass; T2WI scan showing an isointense to hyperintense mass. The arrowheads indicate the tumor; the arrow indicates the retromandibular vein.
- C: Ultrasonography of the right parotid gland showing a 5-cm heterogeneous hypoechoic mass with an acoustic shadow.
- D: FDG-PET-CT scan showing accumulation in the right parotid tumor (SUVmax, 7.27). No cervical lymph node metastasis or distant metastasis was observed.

lier. He felt no pain and had no facial palsy. He had a clinical history of hypertension, diabetes mellitus, hyperlipidemia, and hyperuricemia. Physical examination showed that the right parotid mass (diameter, 5 cm) firmly adhered to adjacent skin. Laboratory analysis of blood biochemistry revealed a blood urea nitrogen level of 42.2 mg/dL, a creatinine (Cr) level of 2.07 mg/dL, and an HbA1c level of 8.0%, indicating renal dysfunction caused by diabetes mellitus.

Plain computed tomography (CT) scans revealed a nearly isodense mass in the right parotid gland (Fig. 1A).

Magnetic resonance imaging (MRI) showed that the mass was heterogeneous, hypointense to isointense on T1WI scans, and isointense to hyperintense on T2WI scans. MRI also showed that the tumor extended beyond the parotid capsule into the superficial fascia and subcutaneous tissue but did not extend deeper than the retromandibular vein, which suggested that it was located superficial to the facial nerve (Fig. 1B). Ultrasonography showed an irregularly edged hypoechoic mass on the right parotid gland (Fig. 1C). Fine-needle aspiration cytology suggested a malignant tumor. Positron emission

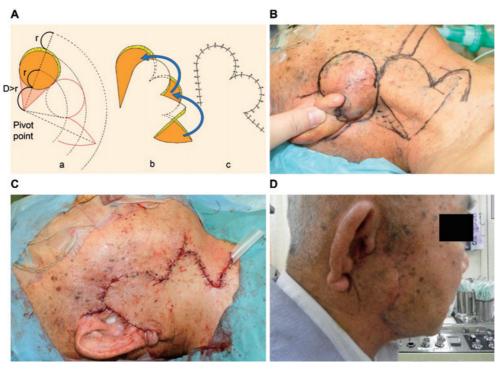


Fig. 2

A: Illustration of the bilobed flap. We designed the bilobed flap to be caudal to the defect. The first lobe was as large as the defect, and the second lobe was designed to be longer and narrower than the first lobe. Both lobes were deployed around the pivot point, the distance of which from the defect edge was longer than its radius.

B: The incision line before resection. The pivot point was set at the lateral side of the flap.

C: Closure of the defect with the bilobed flap.

D: Photograph of the surgical site at 6 months postoperatively.

tomography-CT (PET-CT) revealed local accumulation of fluorodeoxyglucose (FDG), with a standard uptake value max of 7.27 (**Fig. 1D**). Taken together, these findings indicated a diagnosis of parotid carcinoma, cT4aN0M0.

We performed a superficial parotidectomy with excision of the involved skin, which was approximately 6 cm in diameter, and reconstruction using a bilobed flap. To secure a safety margin for resecting the tumor invading the subcutaneous tissue, we outlined the skin incision as a round shape, 6 cm in diameter, and customized the design of the bilobed flap so that it would be caudal to the defect. The first lobe was as large as the defect, and the second lobe was designed to be longer and narrower than the first lobe. Both lobes were deployed around the pivot point, the distance of which from the edge of the defect was longer than its radius (Fig. 2A, B). To avoid overlapping the flap, another skin incision was made to expand the surgical field. After resecting the tumor while preserving the facial nerve, we elevated the bilobed flap and primarily closed the defect (Fig. 2C). A Penrose drain was placed under the flap. There was postoperative facial nerve palsy (26/40 points by the Yanagihara

method; grade 3 by the House-Brackmann method), which completely resolved after 6 months. Postoperative wound color match was good (Fig. 2D).

Histopathological findings of the resected specimens showed mixed areas of salivary duct carcinoma-like lesion with comedonecrosis and sarcoma-like lesions in a low-power view. Tumor invasion of the parotid capsule (blue arrow) and subcutaneous tissue (yellow arrow) were observed (Fig. 3A, B). The tumor was classified as pT4a, as it showed subcutaneous tissue extension. A high-power view revealed a carcinoma-like lesion with sporadic growth of atypical cells and enlargement of the nucleus, suggesting salivary duct carcinoma-like foci. In the surrounding stroma, spindle-shaped sarcoma-like atypical cells were observed (Fig. 4A, B). Immunohistochemical staining showed that the salivary duct carcinoma-like lesion was positive for cytokeratin AE1/ AE3 (Fig. 4C). Some sarcoma-like atypical cells were positive for cytokeratin AE1/AE3. In addition, there was an area where the carcinoma-like area appeared to transition to a sarcoma-like lesion (Fig. 4D). Therefore, we diagnosed sarcomatoid salivary duct carcinoma (SSDC). Al-

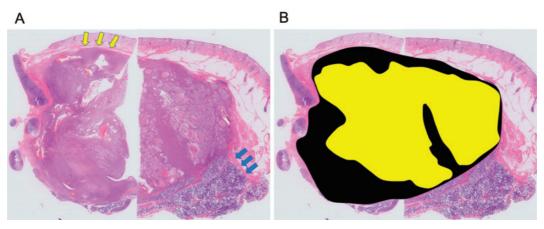


Fig. 3 Histopathological findings: low-power view

A: There were mixed areas of salivary duct carcinoma-like lesion with comedonecrosis and sarcoma-like lesions. Tumor invasion of the parotid capsule (blue arrow) and subcutaneous tissue (yellow arrow) was observed.

B: The ratio of salivary duct carcinoma-like lesions (yellow-filled areas) to sarcoma-like lesions (black-filled areas) was approximately 2:1.

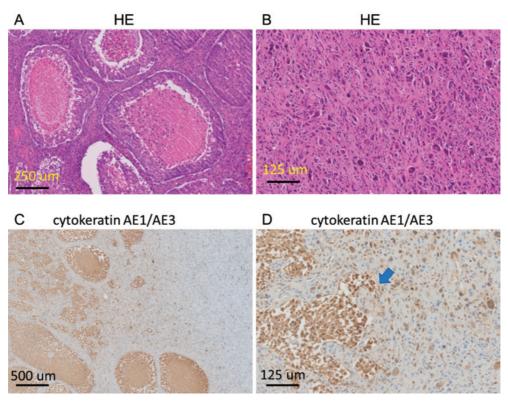


Fig. 4 Histopathological findings: high-power view

A: Histopathological findings of the resected specimens showed sporadic growth of atypical cells and enlargement of the nucleus, suggesting salivary duct carcinoma-like foci.

B: In the surrounding stroma, pleomorphic or spindle-shaped sarcoma-like atypical cells were observed.

C: Immunohistochemical staining showed that the salivary duct carcinoma-like lesion was positive for cytokeratin AE1/AE3.

D: Some sarcoma-like atypical cells were positive for cytokeratin AE1/AE3. In addition, there was an area where the carcinoma-like area appeared to transition to a sarcoma-like lesion (arrow).

though the surgical margin was negative, postoperative irradiation was performed (66 Gy) because the malignancy was high grade. No recurrence of cancer had been observed when the patient died of acute heart failure, 2.5 years after the start of treatment.

Discussion

To our knowledge, this is the first case report to describe the use of a bilobed flap for reconstruction after excision of parotid carcinoma. Reconstruction methods consist of local flaps harvested from adjacent skin and subcutaneous tissue, pedicled flaps (ie, axial-patterned (myo-)cutaneous flaps transposed around the pivot point), and free flaps. Local flaps are classified into a random blood flow pattern, and flap length must not exceed 1.5 to 3 times the width. There is also a report of reconstruction with a rhomboid flap after parotid surgery3. Most pedicled flaps have an axial-patterned vascular pedicle, and it is possible to design an elongated flap or a flap including muscle bodies. The pectoralis major myocutaneous flap, DP flap, latissimus dorsi flap, and supraclavicular flap4 are frequently used. However, for relatively small defects, the submental island flap⁵ and infrahyoid myocutaneous flap, which can be elevated from the neck, can be used. As for free flaps, the fibular flap or scapular flap may be used in cases of segmental mandibulectomy. Among the three flap types, the local flap is the least flexible but is the easiest, least time-consuming, and least invasive procedure. In some cases, the local flap may be more flexible than the pedicled flap, as it does not require consideration of the vascular pedicle. Moreover, even if neck dissection is required, the local flap is not affected. It can be elevated below the subplatysmal layer, unless no skin infiltration of metastatic lymph nodes is present at the donor site. However, if there is a history of neck dissection or irradiation, blood flow to the cervical skin is often poor. A local flap is not indicated in such cases, and a pedicled flap may be better. The pectoralis major myocutaneous flap exhibits less vascular anatomic variation and contains a blood-flow-rich muscle body, making it easier to accommodate relatively large defects. When combined resection of the mandible is performed, a local flap is not indicated because a flap with volume including muscle and bone is required. In such cases, reconstruction using a free composite flap, such as a vascularized, free fibular, osteocutaneous flap, is indicated. In light of these previous findings, a local flap was deemed the most appropriate for our patient, as he was older than 80 years and had complications of diabetic nephropathy.

The bilobed flap is a local flap first reported by Essere in 1918 and is used for reconstructing small defects after excision of nasal tumors. He designed two lobes: one rotated 90° and the other rotated 180° from the defect. With two lobes, the flap can be widened to stabilize blood flow. By shifting the two lobes individually, flap tension can be reduced. In 1989, Zitelli⁶ reported a modification that reduces flap rotation from 180° to between 90° and 110°. This modification allows the flap to be used for larger defects. When used for skin defects in the parotid area, the maximum diameter of the defect that can be accommodated is 6 cm, because of possible dead space when the donor site is caudal to the clavicle.

Although it is difficult to identify nerve invasion on imaging findings such as MRI scans, when considering postoperative function it is standard practice to preserve as much nerve as possible when there is no preoperative facial paralysis and intraoperative findings show no adhesion between the tumor and facial nerve⁷. Our patient had no facial nerve palsy before surgery, and intraoperative findings showed no adhesions to the nerve, so the nerve was preserved.

SSDC is a rare variant of salivary duct carcinoma and was first reported by Henry et al. in 2000⁸. On pathological analysis this biphasic tumor has both a sarcomatoid component and carcinomatous component. In elderly adults, SSDC develops most frequently in the parotid gland, although there have been reports of SSDC in submandibular glands⁹ and minor salivary glands¹⁰. SSDC is rare, and information on its characteristics and prognosis is limited; however, like conventional SDC, it is a high-grade malignant tumor with a poor prognosis. Our patient was old, and we decided to perform postoperative irradiation because of the aggressive nature of this tumor. Unfortunately, he died of acute heart failure at 2.5 years postoperatively.

This case report cannot answer questions related to factors affecting the applicability of the bilobed flap, such as age (which may affect skin elasticity), extent of the defect, effect of neck dissection, and position of the pivot point (medial or lateral to the flap). Thus, further studies with more patients are warranted.

Conclusion

The bilobed flap is the simplest, quickest, and least invasive method for covering a skin defect with a diameter of 6 cm or less in patients with parotid carcinoma. It is especially suitable for elderly patients with comorbidities.

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Declarations: Because the patient died, informed consent to use the photographs in this article was provided by the patient's daughter. The authors obtained consent for publication.

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Conflict of Interest: None declared.

References

- 1. Yenidunya MO, Demirseren ME, Ceran C. Bilobed flap reconstruction in infraorbital skin defects. Plast Reconstr Surg. 2007 Jan;119(1):145–50.
- Zwierz A, Masna K, Burduk P. The use of giant bilobed flaps for tissue defect reconstruction after temporal bone and ear resection. Ear Nose Throat J. 2020 Apr 17; 145561320918986.
- 3. Hung MH, Liao CT, Kang CJ, Huang SF. Local rhomboid flap reconstruction for skin defects after excising large parotid gland tumors. J Oral Maxillofac Surg. 2017 Jan;75(1):

225.e1-.e5.

- Emerick KS, Herr MW, Lin DT, Santos F, Deschler DG. Supraclavicular artery island flap for reconstruction of complex parotidectomy, lateral skull base, and total auriculectomy defects. JAMA Otolaryngol Head Neck Surg. 2014 Sep;140(9):861–6.
- Patel AV, Thuener JE, Clancy K, Ascha M, Manzoor NF, Zender CA. Submental artery island flap versus free flap reconstruction of lateral facial soft tissue and parotidectomy defects: Comparison of outcomes and patient factors. Oral Oncol. 2018 Mar;78:194–9.
- Zitelli JA. The bilobed flap for nasal reconstruction. Arch Dermatol. 1989 Jul;125(7):957–9.
- Guntinas-Lichius O, Silver CE, Thielker J, et al. Management of the facial nerve in parotid cancer: preservation or resection and reconstruction. Eur Arch Otorhinolaryngol. 2018 Nov;275(11):2615–26.
- 8. Henley JD, Seo IS, Dayan D, Gnepp DR. Sarcomatoid salivary duct carcinoma of the parotid gland. Hum Pathol. 2000 Feb;31(2):208–13.
- 9. Nagao T, Gaffey TA, Serizawa H, et al. Sarcomatoid variant of salivary duct carcinoma: clinicopathologic and immunohistochemical study of eight cases with review of the literature. Am J Clin Pathol. 2004 Aug;122(2):222–31.
- 10. Tomihara K, Hamashima T, Nagao T, Nakamori K, Sasahara M, Noguchi M. Sarcomatoid salivary duct carcinoma of the palate: a rare case report. Oral Surg Oral Med Oral Pathol Oral Radiol. 2015 Jan;119(1):e27–32.

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