Bilobed flap for reconstruction of skin defect after excision of parotid carcinoma: a case report

Running title: Parotid reconstruction by bilobed flap

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Author Contributions

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Declarations

Informed consent to use the photographs for publishing in the article was provided by the patient's daughter because the patient was already deceased. The co-authors obtained consent for publication.

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Reconstruction of skin defect after excision of parotid carcinoma: a case report

Running title: Parotid reconstruction by bilobed flap

Abstract

Objectives: Reconstruction of medium-sized skin defect after excision of parotid carcinoma is performed using local flaps, pedicled flaps, or free flaps. The bilobed flap is one of the local flaps adopted by plastic surgeons primarily for a small nasal skin defect. We report a case of a patient with parotid carcinoma with skin infiltration who successfully underwent skin reconstruction with the bilobed flap. **Methods:** An 84-year-old man had visited our hospital with a parotid mass 2 months earlier. He was diagnosed with parotid carcinoma with skin infiltration and underwent radical surgery. The skin defect was round-shaped with a 6 cm diameter. It was resected and reconstructed using a bilobed flap designed caudal to the defect. **Results:** Except for the postoperative facial nerve palsy, which improved within 6 months, the postoperative course was uneventful, and the patient was discharged on the seventh postoperative day. Pathological examination revealed a sarcomatoid salivary duct carcinoma. **Conclusions:** The bilobed flap is useful for reconstructing skin defects measuring up to 6 cm in diameter.

Keywords: Bilobed flap, Parotid grand malignancy, Local flap, Sarcomatoid salivary duct carcinoma, Geriatrics

Introduction

Parotid carcinoma tends to invade surrounding structures such as the subcutaneous tissue, skin, cartilage of the external auditory canal, mandibular bone, and facial nerve. In the case of skin invasion, it is necessary to excise the involved skin. However, stretched skin due to a slow-growing tumor or small defect is closed by reefing with the remaining skin. Nevertheless, patients with a high-grade malignant parotid tumor with rapid growth and direct skin involvement whose defect cannot be closed require skin reconstruction. The skin reconstruction methods comprise local flaps, pedicled flaps, and free flaps. It is necessary to adopt the appropriate flap considering the site and range of the defect and the patient's general condition, including age and comorbidity. The literature reports reconstruction using bilobed flaps for skin defects after tumor resection in the infraorbital, ear, and temporal regions^{1, 2}. However, to the best of our knowledge, no studies have reported using a bilobed flap for reconstruction after excision of parotid carcinoma. Herein, we report a case of an elderly patient with parotid carcinoma who underwent skin excision and a successful reconstruction using the 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 earlier. He felt no pain and had no facial palsy. He had clinical histories of hypertension, diabetes mellitus, hyperlipidemia, and hyperuricemia. Physical examination showed that the right parotid mass had stuck firmly to the adjacent skin with a diameter of 5 cm.

A blood biochemical examination 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) imaging revealed an almost isodense mass in the right parotid gland (Fig. 1A). Magnetic resonance imaging (MRI) showed that the mass was heterogeneous and also demonstrated low- to iso-intensity on T1WI and iso- to high-intensity on T2WI images. 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 the tumor located superficial to the facial nerve. (Fig. 1B). Ultrasonography disclosed an irregularly edged hypoechoic mass on the right parotid gland (Fig. 1C). Fine needle aspiration cytology suggested a malignant tumor without further pathological information. Positron emission tomography-CT (PET-CT) revealed local accumulation of fluorodeoxyglucose (FDG) with a standard uptake value max of 7.27 (Fig. 1D). Accordingly, a diagnosis of parotid carcinoma cT4aN0M0 was made.

We decided to perform a superficial parotidectomy with excision of the involved skin, which was approximately 6 cm in diameter, and reconstruction using the bilobed flap. We outlined the skin incision as a round shape of 6 cm in diameter to secure the safety margin for resecting the tumor invading the subcutaneous tissue. We designed the bilobed flap caudal to the defect. The first lobe was as large as the defect, and the second lobe was designed longer and narrower than the first lobe. Both lobes were deployed around the pivot point whose distance from the edge of the defect was longer than its radius (Fig. 2A, B). Another skin incision was made to expand the surgical field to avoid overlapping the flap. After resecting the tumor while preserving the facial

nerve, we elevated the bilobed flap and primarily closed the defect (Fig. 2C). The Penrose drain was placed under the flap. There was postoperative facial nerve palsy, and the score was 26/40 points as evaluated by the Yanagihara method and grade 3 as assessed by the House-Brackmann method, which completely improved within 6 months.

Histopathological findings of the resected specimens in low-power field showed mixed areas of salivary duct carcinoma-like lesion with comedo necrosis and sarcoma-like lesions. Tumor invasion of parotid capsule (blue arrow) and subcutaneous tissue (yellow arrow) were observed (Fig. 3A, B). The tumor was determined to be pT4a as it showed subcutaneous tissue extension. In high-power field, carcinoma-like lesion showed sporadic growth of atypical cells with enlargement of 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 of the 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, the diagnosis was sarcomatoid salivary duct carcinoma (SSDC). Although the surgical margin was negative, postoperative irradiation was performed with 66 Gy because of high-grade malignancy. The patient died of acute heart failure 2.5 years after the initiation of treatment without recurrence.

Discussion

To our knowledge, this is the first case report of using a bilobed flap for reconstruction after excision of parotid carcinoma. The reconstruction methods consist of local flaps

harvested from the adjacent skin and subcutaneous tissue, pedicled flaps that are axial-patterned (myo-)cutaneous flaps transposed around the pivot point, and free flaps. The local flap is classified into a random blood flow pattern, and the flap length is limited to 1.5–3 times longer than the width. There is also a reconstruction report with the rhomboid flap after parotid surgery³. The pedicled flap primarily has an axial-patterned vascular pedicle, and it is possible to design an elongated flap or a flap including muscular bodies. The major pectoralis flap, DP flap, latissimus dorsi flap, and supraclavicular flap⁴ are frequently used. However, for relatively small defects, the submental island flap⁵ and the infrahyoid myocutaneous flap that can be elevated from the neck are also used. As the free flap, in case of segmental mandibulectomy, the fibular flap or the scapular flap may be used. Among these three flaps, 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 rather 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 by dissection. 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, and in such a case, the local flap is not indicated, and a pedicled flap may be better. The pectoralis major myocutaneous flap (PMMCF) has less vascular anatomy 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.

Based on the above-described information, the local flap was the most appropriate for our patient as he was aged >80 years and had complications of diabetic nephropathy. The bilobed flap is one of the local flaps first reported by Essere in 1918, which is used for the reconstruction of a small defect after excision of the nose tumor. He designed the two lobes in which one lobe is rotated 90°, and another lobe is rotated 180° from the defect. With two lobes, the flap can be made wider to stabilize the blood flow, and by shifting the two lobes one by one, the flap tension can be reduced. In 1989, Zitelli⁶ reported the modification that reduces the flap's rotation from 180° to between 90° and 110°. Due to this modification, the flap can be adopted for a larger defect. 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 exceeds caudal to the clavicle.

Although it is difficult to predict the presence or absence of nerve invasion based on imaging findings such as MRI, it is common practice to preserve the nerve as much as possible if there is no preoperative facial paralysis and intraoperative findings show no adhesion between the tumor and the facial nerve, taking into account the postoperative function⁷. In this case, there was no facial nerve palsy before surgery, and intraoperative findings showed no adhesions to the nerve, so the nerve was preserved.

Concerning SSDC, it is a rare variant of SDC that was first reported by Henry et al. in 2000⁸. The biphasic tumor's pathological finding has both a sarcomatoid component and a carcinomatous component. SSDC develops in elderly patients most frequently in the parotid gland, although there have been reports on SSDC in the submandibular glands⁹ and minor salivary glands¹⁰. SSDC is rare, and there is no extensive information about its nature and prognosis; however, like conventional SDC, it is a high-grade

malignant tumor and has a poor prognosis. Our case was also an elderly patient, and we decided to perform postoperative irradiation for the aggressive nature of this tumor, but he died of acute heart failure after 2.5 years.

As this is a case report, factors affecting the applicability of the bilobed flap, such as age (which may affect skin stretchability), extent of the defect, effect of neck dissection, and position of pivot point (medial or lateral to the flap), remain unknown. Hence, further studies with more cases are warranted.

Conclusion

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

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Declaration of Conflicting Interests

The authors declare that there is no conflict of interest.

References

1. Yenidunya MO, Demirseren ME, Ceran C. Bilobed flap reconstruction in infraorbital skin defects. *Plast Reconstr Surg*. 2007 Jan;119(1):145-50.
2. 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.
4. 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.
5. 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.
6. Zitelli JA. The bilobed flap for nasal reconstruction. *Arch Dermatol*. 1989 Jul;125(7):957-9.
7. 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.

Fig. Legends

Fig. 1.

A: Plane CT axial view revealed an almost isodense mass in the right parotid gland.

B: Plane MRI axial view revealed a mass with low- to iso-intensity on T1WI and iso- to high-intensity on T2WI images in the right parotid gland. The arrowheads indicate the tumor whereas the arrow indicates retromandibular vein.

C: Ultrasonography revealed a 5-cm heterogeneous hypoechoic mass with acoustic shadow in the right parotid gland.

D: FDG-PET-CT showed an accumulation in the right parotid tumor with a SUV_{max} of 7.27. Neither cervical lymph node metastasis nor distant metastasis was observed.

Fig. 2.

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

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

C: Closure of the defect with the bilobed flap.

D: Postoperative appearance at 6 months.

Fig.3. Histopathological findings of low-power field

A: There were mixed areas of salivary duct carcinoma-like lesion with comedo necrosis and sarcoma-like lesions. Tumor invasion of parotid capsule (blue arrow) and subcutaneous tissue (yellow arrow) were 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 of high-power field

A: Histopathological findings of the resected specimens showed sporadic growth of atypical cells with enlargement of 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 of the 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).

Figures

Fig.1.

Fig. 2.

Fig.3.

Fig.4.