

CASE REPORT

Intra-flap recurrence of an oral squamous cell carcinoma after curative resection and reconstruction with a free flap

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Key Clinical Message

Intra-flap recurrence after reconstruction is a possible complication in oral squamous cell carcinoma (OSCC) after curative resection and reconstruction with a free flap.

Abstract

This case report describes an intra-flap recurrence after tumor resection and reconstruction with a latissimus dorsi flap due to OSCC. We report a 58-year-old female patient who underwent curative resection of a squamous cell carcinoma in the oral cavity and submental area and reconstruction with a latissimus dorsi free flap. Thirteen months after the operation, the patient presented with lung metastases and, 22 months after the operation, a metastatic lesion in the flap's central area with no sign of recurrence in the primary resection site occurred. The patient died 25 months after the initial operation due to the rapid progression of the disease and further distant metastases. To our knowledge, similar cases do not exist in the literature. This is the first reported case of intra-flap metastasis after surgical treatment of OSCC.

KEYWORDS

case report, free tissue flap, intra-flap recurrence, neoplasm metastases, squamous cell carcinoma of the head and neck

1 | INTRODUCTION

Oral squamous cell carcinoma (OSCC) is the most common cancer in the oral cavity.¹ The 5-year survival rate amounts to 64.4%–79.3%.² However, the prognosis worsens due to metastases and survival rates potentially decrease by 50%.³ The prognosis is also compromised when local recurrence appears.⁴ In the current literature local recurrence patterns after curative resection and reconstruction

with free flaps were evaluated in oropharyngeal cancers and reported, that the primary site of local recurrence is usually in the vicinity of the anastomosis margin, outside the flap tissue and not in the flap itself.⁵ Free flaps have become a standard method for the reconstruction of defects in the head and neck area.⁶ For reconstruction of defects, soft tissue flaps, osseous transplants or combinations are available. So far, no data exists regarding intra-flap metastases. In this case report, we present a 58-year-old female

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patient with an intra-flap metastasis in a latissimus dorsi flap after resection of an OSCC.

2 | CASE HISTORY AND METHODS

A 58-year-old female with a 1-month history of painless progressive swelling in the front area of the mouth floor was referred to our department. The patient was a heavy smoker with 60 pack years. An intraoral biopsy of the pathology revealed a squamous cell carcinoma. The patient underwent a computed tomography scan (CT scan) for staging, which showed a tumor mass in the anterior floor of the mouth adjacent to the mandible and extending to the skin of the submental area (Figure 1). There was no sign of bone involvement but prominent cervical lymph nodes on both sides. There was no sign of distant disease.

According to the tumor board's decision, surgical intervention was performed. Together with the lingual part of the lower jaw bone and the submental skin, the frontal area of the mouth floor was removed to achieve negative resection margins (Figure 2). In addition, neck dissection of levels I–III on both sides was performed. Intraoperative frozen sections revealed lymph node metastasis in Level III on both sides. Therefore it was decided to perform a modified radical neck dissection according to the German-Evidence-based Guidelines for Oral Cavity Cancer on both sides.

The soft tissue defect was reconstructed microsurgically with a latissimus dorsi myocutaneous transplant from the left side. A tracheotomy was performed to avoid postoperative airway compromise.

Histological examination of the resected tissues showed negative resection margins, and the minimum margin

distance was 5 mm in the anterior part of the tongue. The margins are described as following in the histopathological report: Medial: 0.9 cm lateral: 2.2 cm, basal: 0.7 cm, anterior: 1.0 cm, posterior: 0.8 cm. There was no perineural infiltration found. Infiltration of the submental skin with an absence of bone involvement was also confirmed histologically. Metastases at the regional lymph nodes at level III were reported on both sides (3 out of 81 resected lymph nodes). There was no sign of blood or lymph vessel invasion and no extranodal extension. The tumor was classified as pT4a, pN2c (3/81 lymph nodes, maximum size 12 mm), L1, V0, Pn0, G3, UICC Stage IVa according to TNM-UICC 2017 classification.⁷

The patient received adjuvant radiochemotherapy with cisplatin and carboplatin and 66 gray radiation divided into four cycles. Recurrence was diagnosed during follow-up control 13 months after the operation. A CT scan (Figure 3) followed by a positron emission tomography—computer tomography (PET–CT) scan revealed two suspect pulmonary round foci, one at each side, with enlarged but not highly suspect hilar and mediastinal lymph nodes. Bronchoscopy revealed a pulmonary metastasis of the primary OSCC. The patient received three cycles of chemotherapy with paclitaxel and carboplatin. After the third cycle of chemotherapy, remission of the pulmonary foci was noted, but treatment was interrupted due to side effects such as dysgeusia, dysarthria, trismus, dysphagia, dysphonia, and mucositis.

3 | RESULTS

Twenty-two months after the initial operation and during remission of the lung metastases, a CT scan of the neck revealed a suspicious cutaneous round lesion

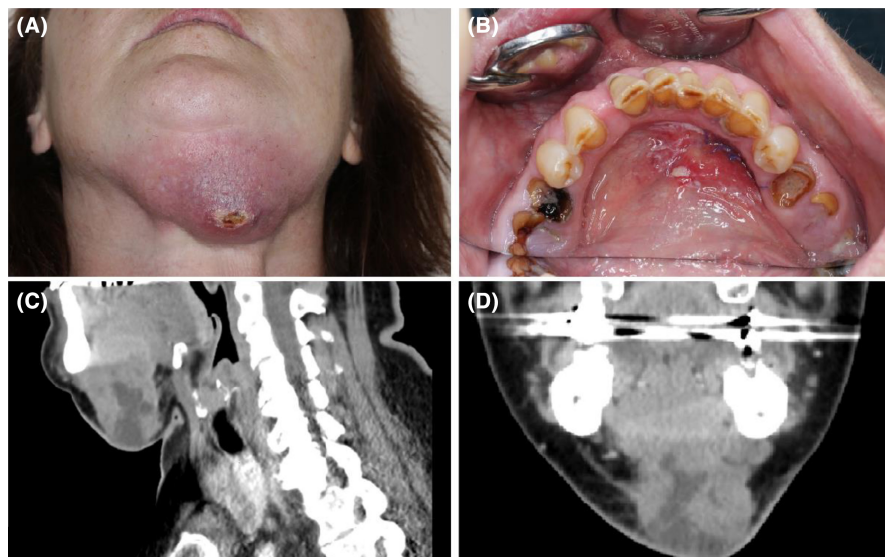


FIGURE 1 Preoperative photos and CT scan. (A, B) Clinical examination. (C, D) sagittal, coronal CT scan showing necrotic hypodense tumor with peripheral rim enhancement.

FIGURE 2 Intraoperative photos. (A, B) showing the intraoperative situs with inserted osteosynthesis plate and latissimus dorsi myocutaneous transplant for reconstructing soft tissue defects of the floor of mouth.

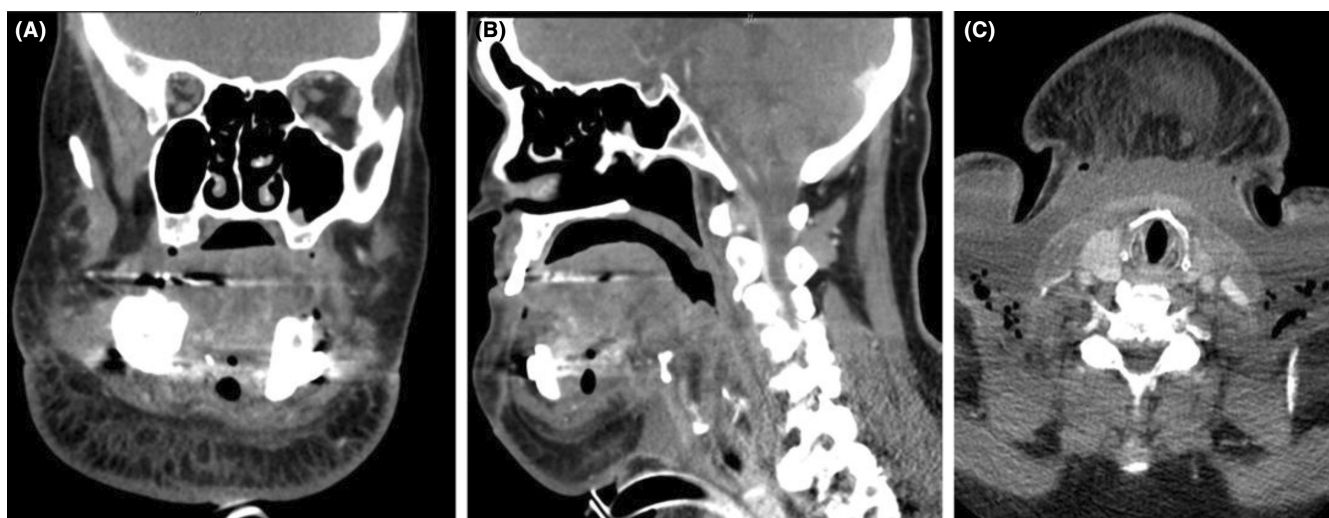
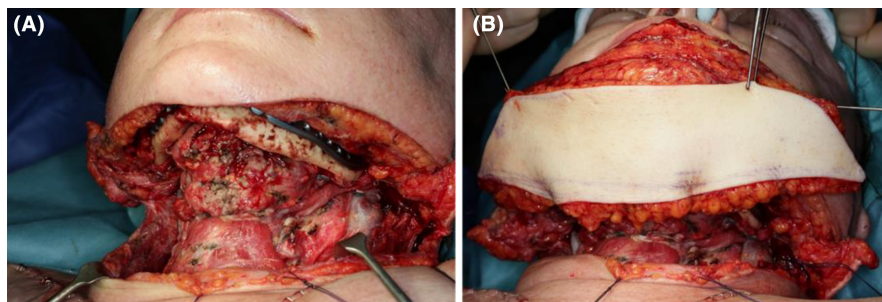


FIGURE 3 Postoperative CT scan before appearance of the local recurrence (14 months before appearance of intraflap metastases). (A–C) Coronal, sagittal, axial scans showing subcutaneous fat oedema, and air bubbles within supra clavicular fossae (early postoperative CT scan).

(13.4×16.4×15.6 mm) in the central area of the latissimus dorsi flap in the submental area (Figure 4C–E). Erythema was present in the overlying skin (Figure 4A,B). An excisional biopsy with wide margins revealed a squamous cell carcinoma with the same characteristics as the resected primary oral cancer. The metastasis was removed with negative margins.

Two months after the metastasectomy of the free flap's central area, the disease progressed rapidly with further lung, bone, and brain metastases but no further local recurrence. The patient deceased 25 months after the initial operation and 3 months after the resection of the recurrent tumor.

4 | DISCUSSION

Studies examining the sites of local recurrence, following flap reconstruction in patients with oropharyngeal cancers, are very limited in current literature. Cho et al.⁵ reported that the primary site of local recurrence was in the vicinity of the anastomosis margin outside the flap

tissue. Out of 114 patients studied, no subject presented with a recurrent tumor in the flap, which has important implications on the radiotherapy target volumes to reduce postoperative morbidities as radiation-associated morbidity.⁸ In addition, Ellison et al. described recurrent tumors in the base of the flap pedicle following cancer resection and myocutaneous flap reconstruction in three cases.⁹ The purpose of this case report is to raise awareness of the possibility of tumor recurrence in reconstruction flaps to facilitate early diagnosis and aggressive treatment.

In our case, the intra-flap recurrent tumor is located approximately 1.4 cm from the inferior margin of the latissimus dorsi musculocutaneous flap, with the erythema located adjacent to this margin. A possible explanation that needs to be considered is the intra-flap recurrence as a distant metastasis located in the flap. Still, iatrogenic implantation of exfoliated malignant cells could be suggested as a possible mechanism of metastasis as well.¹⁰ However, the uncommon site (central area of the flap) of the tumor tissue, the site's irrigation after the closure of the resection field, and the lack of known intraoperative or postoperative injuries or wounds in the flap tissue do

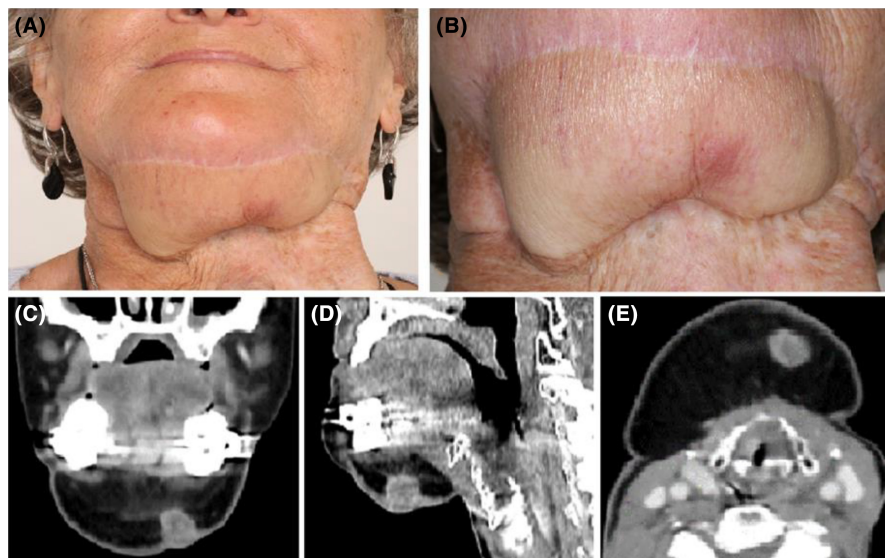


FIGURE 4 Photos and CT scan after the appearance of the local recurrence in the flap. (A, B) showing the clinical and (C–E) showing the CT appearance of the numular recurrence in the flap.

not fully support this mechanism. Anyway, the erythema that touched the inferior flap margin could indicate the origin of the recurrent tumor in the resection margin.

A second possibility could be the presence of a cutaneous metastasis at the donor site prior to surgical removal of the transplant. In our case, neither the clinical examination nor the CT scan showed signs of cutaneous metastasis in the donor site. Another possible mechanism of recurrence could be the endophytic ingrowth from the primary poorly differentiated (G3) tumor location (anterior floor of the mouth or sublingual area) despite the initially confirmed R0-resection. Figure 3D presents a spidery contrast agent enhancement cranially to the intra-flap enlargement. Clinical and radiological assessments are difficult and may not fully be reliable (see artifact caused by the metallic osteosynthesis plate). It is also known, that titanium osteosynthesis material leads to a dose reduction (8.5%, compared to bone 2.2%) due to a scattering of therapeutic radiation.¹¹

After the evaluation of the radiological and clinical parameters, the bilateral pulmonary nodules were considered metastases of the resected oral carcinoma and not metachronous, multifocal lung cancer. However, it is often difficult to discern a lung nodule's nature using standard clinical and histologic parameters.¹² In our case, further examinations were not performed due to the patient's known history.¹³ The lung tumor masses were preexisting for 9 months before the intra-flap lesion was detected and there was clinical and radiological remission of the disease for 9 months after the lung oligometastases were diagnosed. Therefore cutaneous metastasis originating from the pulmonary masses is unlikely.

Radical, local treatment through surgery or radiotherapy for an oligometastatic disease is a recognized therapeutic modality for treating metachronous metastatic disease.¹⁴ Florescu et al. report a 20% 5-year survival rate

in selected patients after pulmonary/liver metastectomy. They also report a 35% 2-year survival rate after stereotactic ablative radiotherapy, which could be efficient not only for lung or liver oligometastases but bone and other locations, too.¹⁵

We experienced a rare case of local recurrence that occurred in the central area of the free flap. Before this intra-flap lesion, there was no sign of recurrence in the resection site, but lung metastases in remission. Although the recurrence mechanisms cannot be clarified with certainty, a resection of the recurrent tumor with wide margins is indicated to remove and examine it. The patterns of local recurrence are of great importance, as tumor seeding at the time of cancer resection as well as hematogenous dissemination need to be considered. We therefore, present this unique case to raise awareness of the possibility of tumor involvement in reconstructive flaps to facilitate early diagnosis and aggressive treatment.

5 | CONCLUSION

Intra-flap recurrence after reconstruction is a possible complication in OSCC after curative resection and reconstruction with a free flap.

AUTHOR CONTRIBUTIONS

Paris Liokatis: Conceptualization; visualization; writing – original draft. **Ioanna Liokati:** Methodology; writing – original draft. **Florian Fegg:** Resources; writing – original draft. **Ina Dewenter:** Resources; visualization; writing – review and editing. **Gerson Mast:** Methodology; supervision; writing – review and editing. **Sven Otto:** Methodology; supervision; writing – review and editing. **Katharina Theresa Obermeier:** Conceptualization; project administration; writing – original draft.

CONFLICT OF INTEREST STATEMENT

The author(s) declare that they have no competing interests.

DATA AVAILABILITY STATEMENT

Data available on request due to privacy.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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