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Case Report

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Two Extremely Rare Cases of Oral Cancer Recurrence in Pectoralis Major Donor Site

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ABSTRACT

Globally, a vast number of oral cancers are treated with resection and immediate reconstruction. This report presents two rare cases of oral cancer recurrence at the pectoralis major flap donor site, with the conventional idea of "cancer seeding" into a potential soil. Despite the rarity, these cases suggest the need for further research into oral cancer subtypes that may disseminate through seeding. While the clinical significance may be debated, the possibility of seeding and the affinity of metastatic clones to donor site tissues should not be ignored. Although molecular understanding of oral cancer metastasis has advanced, such cases remain a scientific conundrum.

Keywords: Oral Cancer, Pectoralis Major Flap, Cancer Seeding

INTRODUCTION

Despite increased understanding of cancer biology and advances in the treatment of oral cancer, recurrence rates remain high in advanced cases, ultimately resulting in poor survival and prognosis. The majority of recurrences occur in locoregional tissues, with a comparatively low number of distant metastases, such as those in the liver, lungs, and distant bones. Flap donor site recurrences for oral cancer are extremely rare, although there are reports of recurrences in regional flap sites and distant free flap donor sites (1).

There are several theories proposed for cancer metastasis. In 1889, Stephen Paget proposed a hypothesis widely known as the “Seed and Soil” concept. The essence of this concept is that “only fertile soil can grasp the metastatic seeds from the tumour.” Later, James Ewing proposed a theory of “mechanical mechanisms,” which explains that only anatomy and vascularity will determine where metastatic cells will deposit. Novel concepts discuss the “tumour self-seeding” hypothesis, as well as the concepts of “pre-metastatic niche” and “dormant niche.” The tumour self-seeding hypothesis explains that circulating tumour cells can lodge in regional or distant organs and can also seed into the primary tumour itself. The pre-metastatic niche concept explains how the environment becomes conducive to harbouring the metastatic cells. The dormant niche concept is introduced to explain the time lapse between the spread of the seeds and metastatic outgrowth. During this time gap, metastatic cells remain dormant as quiescent cells. Current understanding

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of oral cancer metastasis is a combination of all these theories (2).

CASE 01

A 38-year-old male patient presented to a District General Hospital with ipsilateral neck failure following treatment for T3N0M0 primary lateral tongue squamous cell carcinoma, six months after his initial surgery. There was no local recurrence in the tongue, but multiple large neck nodes were identified, and contrast-enhanced computed tomography suggested extracapsular spread of recurrent tumour in the neck. At the time of operation, the condition was categorized as rT3N3M0. The patient underwent salvage radical neck dissection. The neck defect was covered with a pectoralis major myocutaneous (PMMC) flap from the same side. Before moving on to harvesting the flap, neck site was copiously irrigated with distilled water. Gloves changed separating the cancer site and the donor site. During the harvest of the flap, separate instruments were used with maximum precautions to prevent cross contamination of neck and donor site. Excision histology revealed involved and close margins with multiple extracapsular nodes, prompting the patient to undergo chemotherapy and radiation postoperatively with curative intent. He presented to the clinic two months after completing chemotherapy and radiation with recurrences in the superior margin of the flap in the neck. A peculiar swelling was observed in the chest below the clavicle, located at the donor site base of the pedicle (Figure 01). Fine needle aspiration cytology revealed it to be a metastatic deposit of squamous cell carcinoma. He was treated with palliative intent and passed away two months later.

CASE 02

A 56-year-old patient underwent surgery for left buccal mucosal/lower alveolar ridge squamous cell carcinoma. The treatment involved primary site resection, selective neck dissection, and pectoralis major myocutaneous flap reconstruction of the defect. Universal precautions taken to keep any cross contamination between cancer sites and donor site. Excision histology revealed the tumour to be T4N1M0. The patient received adjuvant radiotherapy as indicated by the excision histopathology. He presented to an Oral and Maxillofacial Clinic at a University Dental Hospital, 10 months after completing adjuvant therapy. A prominent lump was observed at the pectoralis major donor site, which was found to be metastatic squamous cell carcinoma via fine needle aspiration biopsy (Figure 02). He showed no recurrence at the primary site. The patient underwent excision of the recurrence and was followed up with no further evidence of disease.

Figure 01: Patient in case 01 having base of the pectoralis major myocutaneous flap pedicle-pointed in red arrow

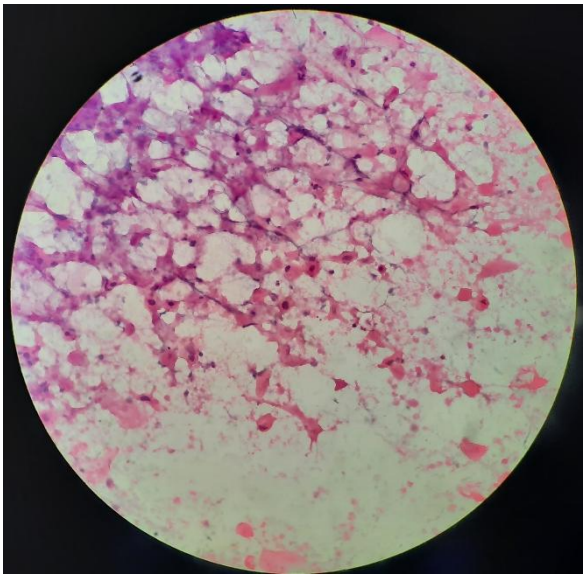

Figure 02: Case 02 patient, Photo microscopy of fine needle aspiration showing evidence of atypical squamous cells, which is suggestive of metastatic squamous cell carcinoma

DISCUSSION

Flap donor site recurrences of oral squamous cell carcinoma reported in the literature are very rare. There are reports of oral cancer recurrence at the pectoralis major donor site, free flap donor site, tracheostomy scar site, and percutaneous gastrostomy insertion site. The following table summarizes the reports of pectoralis major flap donor site recurrences.

The two currently reported cases are similar to those described by the authors mentioned above (Table 01). The review indicates that the majority of patients were young and presented with neck positivity and advanced disease. All recurrences were observed within 12 months of completing treatment for oral cancer, highlighting the aggressive behaviour of these tumors. In addition

Table 01: Literature summary of PMMC flap donor site recurrence of oral cancer

References of the case reports	Case history
1988 Badellino et al.(3)	A-37-year-old male, with T4N1M0 OSCC of floor of the mouth. Treated with surgery, radiotherapy and chemotherapy. Presented with PMMC flap vascular pedicle base recurrence together with primary site recurrence after 12 months.
1994 Stephen S. Kroll et al.(4)	A 39-year-old male with T4N2 Laryngeal squamous cell carcinoma underwent surgery and PMMC flap reconstruction. It was observed recurrence in the chest wall after 3 months.
2001 M Jog et Al.(5)	A 53-year-old male with T3N1M0 right tonsillar cancer extending to retromolar trigone treated with surgery and adjuvant radiotherapy. Presented with only PMMC flap donor site recurrence after 12 months.
2001 M Jog et al.(5)	A 61-year-old male with T3N1M0 tongue cancer (which involved base of the tongue) was treated with surgery and adjuvant radiotherapy. He presented to the clinic with only PMMC base of vascular pedicle recurrence after 8 months.
2004 Bansal et al.(6)	A 45-year-old male, with primary stage III (Neck positive with extracapsular spread) left lower alveolus squamous cell carcinoma which was treated with surgery and adjuvant radiotherapy. He came up with a donor site recurrence after 2 months post treatment with no signs of recurrence in primary site.
2009 Senniappan Kartikeyan et al.(7)	A 35-year-old female with anterior tongue squamous cell carcinoma with involvement of floor of the mouth was treated with surgery (T4N2Mx) and adjuvant radiotherapy. Presented with recurrence in donor site and primary site after 6 months post treatment.
2013 Vasu Reddy C et al.(8)	A 35-year-old male with right buccal mucosal squamous cell carcinoma (advanced stage) treated with surgery, chemotherapy and radiotherapy. He presented with PMMC flap donor site recurrence after 2 months.
2018 R. Kein et al.(9)	A 50-year-old male with right floor of the mouth squamous cell carcinoma treated with surgery and radiotherapy, Developed a recurrence in the PMMC flap donor site after 3 months from the surgery,
2021 P Chandrapattan et al(9)	A 46-year-old male treated for left retromolar squamous cell carcinoma with surgery and radiotherapy with PMMC flap reconstruction. He was disease free for 3 years, recurrence was seen in the same site with neck positivity on fourth year. Again, treated with surgery. He developed recurrence in the PMMC flap donor site after 11 months from recurrent cancer surgery.

to these reports, there are instances of free flap donor site seeding with oral squamous cell carcinoma (10).

Explaining the exact phenomenon of metastasis in these situations is challenging. One possibility is that tumour cells detached from the neck, seeded in the donor bed before the site was closed, even though maximum effort are taken to prevent any cross contamination between sites. Another alternative explanation could be hematogenous or vascular spread of the tumour cells along the flap's pedicle.

It is fascinating to consider the conventional concept of seeding during surgery, as it would require a detached clump of cells to enter a healthy donor site, survive the host immune response, adhere to the tissues while maintaining viability, and then grow further. One could argue that this could involve tumour stem cell seeding, which has the potential to differentiate into different clones within the donor tissues. Further in vitro studies are required for better understanding of the exact mechanisms involved.

As a take-home message for oral and maxillofacial surgeons, it is of utmost importance to prevent contamination of the donor site from the primary cancer site. One could argue that the implantation of tumours in these cases was iatrogenic.

Author declaration

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