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Tumor Recurrence at the Tracheostomy Stoma Following Oncologic Surgery for Squamous Cell Carcinoma of the Tongue: A Case Report and Literature Review

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Abstract

Tumor recurrence at the stoma of a temporary tracheostomy is an uncommon but serious complication of surgical treatment for head and neck squamous cell carcinoma. We report the case of a 52-year-old farmer, former smoker (3.75 pack-years), with tongue squamous cell carcinoma initially managed at another institution with two partial glossectomies and an oncologically insufficient result. He underwent compartmental glossectomy, bilateral modified radical neck dissection, tracheostomy performed before tumor resection, and microvascular reconstruction with an anterolateral thigh flap; pathology confirmed pT4aN0M0 with negative margins and no metastatic involvement in any of the 91 examined lymph nodes, and he received adjuvant radiotherapy without concomitant chemotherapy. During the last week of adjuvant radiotherapy, approximately 16 weeks after surgery, a rapidly progressive vegetant lesion appeared around the tracheostoma; biopsy confirmed poorly differentiated squamous cell carcinoma recurrence at that site, not amenable to surgical salvage due to the extent of the disease and involvement of adjacent structures. The patient subsequently developed tracheitis, aspiration pneumonia, and tumor progression, with severe functional decline and exclusive palliative care until death. Notably, this recurrence occurred in the absence of histopathologically demonstrable nodal disease and became clinically evident during radiotherapy. Mechanical tumor seeding is a plausible explanatory mechanism, particularly when tracheostomy precedes tumor resection, although a definitive causal relationship could not be established. Any vegetant, indurated, ulcerated, or hemorrhagic lesion arising at a tracheostomy stoma in a patient with a history of head and neck squamous cell carcinoma should prompt targeted oncologic evaluation and, when appropriate, histopathological confirmation. Whether considering the anatomic relationship between the tracheostomy site and the irradiation volumes in selected patients can reduce this risk remains to be determined. Further studies are needed to clarify the true role of tumor implantation and to determine whether specific modifications in surgical technique or radiotherapy planning can reduce the risk of this rare but serious complication.

Categories: Otolaryngology, General Surgery, Oncology
Keywords: glossectomy, head and neck neoplasms, neoplasm recurrence local, neoplasm seeding, squamous cell carcinoma (scc), tongue neoplasms, tracheostomy

Introduction

Squamous cell carcinoma is the predominant histological type among malignant neoplasms of the oral cavity, with the tongue representing one of the most frequently affected subsites. Surgical resection of the primary tumor with histologically negative margins remains the treatment of choice: margin status - not merely its reported negativity on pathology, but its actual width - has been described as an essential independent factor for local control and disease-free survival in tongue squamous cell carcinoma [1].

In advanced oral cavity neoplasms involving structures such as the tongue base, oncologic surgery is frequently accompanied by concomitant procedures (neck dissection, gastrostomy, and, often, tracheostomy) aimed at securing the airway and ensuring nutrition during the immediate postoperative period. A tracheostomy is a surgical opening created in the anterior neck into the trachea to allow breathing; its external opening is called the stoma. Perioperative tracheostomy, historically performed routinely in these patients, is not free of complications. Current practice therefore favors its selective use, guided by validated risk scores (Cameron Score and TRACHY Score), and reserves it for T3-T4 tumors, midline or tongue base involvement, bilateral neck dissections, and reconstructions with bulky flaps [2].

An uncommon but feared complication of tracheostomy performed in the oncologic setting is tumor recurrence at the tracheal stoma itself, a phenomenon primarily attributed to mechanical seeding (the implantation of viable tumor cells in healthy tissue) during surgical manipulation of the primary tumor [3]. Unlike stomal recurrence following total laryngectomy, where direct anatomical continuity exists between

the tumor and the airway, recurrence at the stoma of a temporary tracheostomy performed for an oral cavity tumor, in which the tumor has no anatomical continuity with the airway, is more difficult to explain and has been interpreted as suggestive of intraoperative seeding [4,5]. Its reported incidence ranges from 1% to 1.2% of patients undergoing temporary tracheostomy for head and neck cancer [4,6], with a generally poor prognosis once established [4]. However, its precise mechanism, the contribution of surgical technique, and the role of postoperative radiotherapy remain uncertain.

This case raises two practical questions: whether the timing of the tracheostomy relative to tumor resection influences the risk of stomal recurrence, and whether the position of the stoma relative to the radiotherapy field does. We report a patient with tongue squamous cell carcinoma who had initially undergone surgery at another institution with an oncologically insufficient outcome and who, after definitive oncologic resection, bilateral neck dissection, and tracheostomy performed at our institution, developed tumor recurrence at the tracheal stoma. We review the proposed pathophysiological mechanisms, the available evidence on its prevention, and the lessons applicable to head and neck oncologic surgical practice.

Case Presentation

We report the case of a 52-year-old man, a farmer from a rural area with a primary-school level of education. Relevant history included prior smoking, with an approximate consumption of 15 cigarettes per day for five years (a 3.75 pack-year index), discontinued years before presentation, and social alcohol consumption. No other pathological, allergic, or traumatic history of relevance was reported. Regarding family history, his father had chronic obstructive pulmonary disease.

The patient was referred to our institution for a lesion of approximately two years' duration, initially located in the anterior third of the tongue, with a histopathological diagnosis of squamous cell carcinoma. He had previously undergone two wide local excisions via partial hemiglossectomy at another institution, performed five months apart, without achieving definitive oncologic control.

Review of the histopathological reports from both resections showed an infiltrating, keratinizing, moderately differentiated, ulcerated large-cell squamous cell carcinoma. In the second resection specimen, the tumor measured 3 cm in its greatest dimension, with a depth of invasion of 16 mm and no evidence of lymphovascular or perineural invasion. The surgical margins were histologically free of tumor, although close, with a minimum distance of less than 1 mm.

On admission to our institution, physical examination revealed an irregular, ulcerated lesion involving the anterior two-thirds of the tongue, associated with tongue fixation and cicatricial retraction. No palpable cervical lymphadenopathy was identified (Figure *1*).

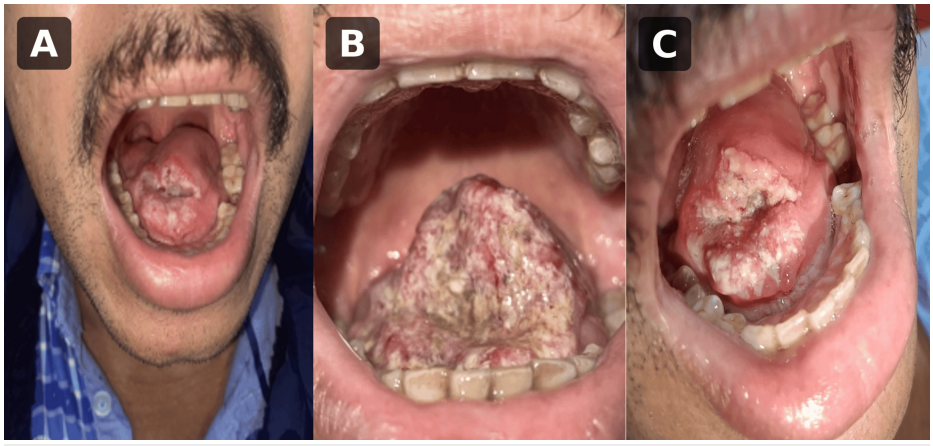


FIGURE 1: Intraoral photographs at admission

(A-C) Irregular, ulcerated lesion of the anterior two-thirds of the tongue with cicatricial retraction and fixation. No palpable cervical lymphadenopathy was observed.

Contrast-enhanced computed tomography of the neck revealed decreased thickness of the anterior two-thirds of the tongue dorsum and edema of the right submandibular gland (Figure *2*). Subsequently, magnetic resonance imaging of the face and neck revealed postsurgical changes secondary to the prior partial glossectomy and a lesion compatible with residual tumor or recurrence, centered on the mucosa and intrinsic musculature of the tongue. The cleavage plane was preserved, with no evidence of infiltration of the sublingual fat, mandible, or submandibular space. No cervical lymphadenopathy suspicious for metastatic involvement was identified (Figure *3*). Contrast-enhanced chest computed tomography revealed

no evidence of pulmonary metastatic disease.

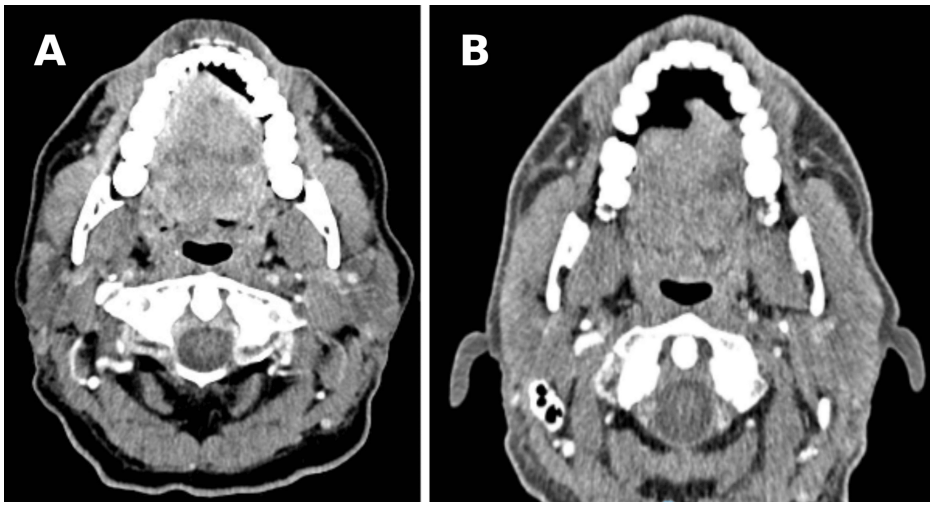


FIGURE 2: Contrast-enhanced neck computed tomography at admission

(A, B) Axial slices from the same study at different levels show decreased thickness in the anterior two-thirds of the tongue dorsum, consistent with physical examination findings.

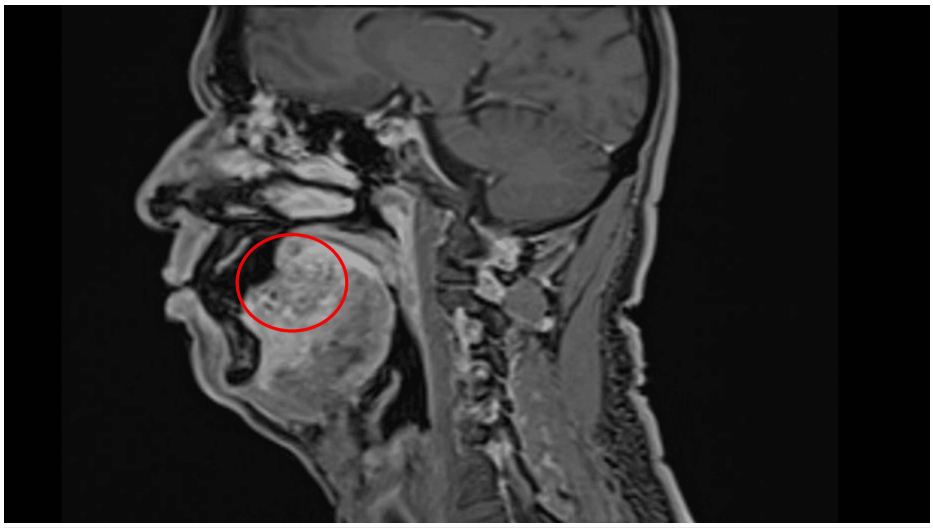


FIGURE 3: Sagittal contrast-enhanced magnetic resonance imaging of the face and neck

Postsurgical changes after partial glossectomy with resection of the anterior third of the tongue. The red circle marks the anterosuperior portion of the tongue, where residual tissue versus tumor recurrence is identified in the surgical bed, without infiltration of the sublingual fat, mandible, or submandibular space. No cervical lymphadenopathy suggestive of secondary neoplastic involvement is observed.

The case was presented at a multidisciplinary tumor board. Based on the clinical and radiological findings, the tumor was staged as cT4aN0M0, clinical stage IVA according to the eighth edition of the American Joint Committee on Cancer (AJCC) staging system [7]. Given the patient's adequate functional status (Eastern Cooperative Oncology Group (ECOG) performance status of 1) and the absence of distant metastatic disease, surgical treatment with oncologic intent was planned, consisting of compartmental glossectomy with bilateral modified radical neck dissection and reconstruction with an anterolateral thigh (ALT) microvascular flap.

The patient underwent surgery, during which the following procedures were performed sequentially: endoscopic gastrostomy, percutaneous tracheostomy with a 7.5-mm cannula, bilateral modified radical neck dissection of levels IA, IIA, IIB, III, IV, and V, and bilateral compartmental glossectomy with en bloc resection via a transcervical "pull-through" approach. The tracheostomy was performed in the midline, 2 cm

above the suprasternal notch, before tumor resection, and the pull-through route was 3 cm from the stoma. The surgical bed was irrigated with 0.9% saline solution during hemostasis control before wound closure. The resulting defect was reconstructed using a free microvascular ALT flap (Figure 4). The immediate postoperative outcomes are shown in Figure 5.

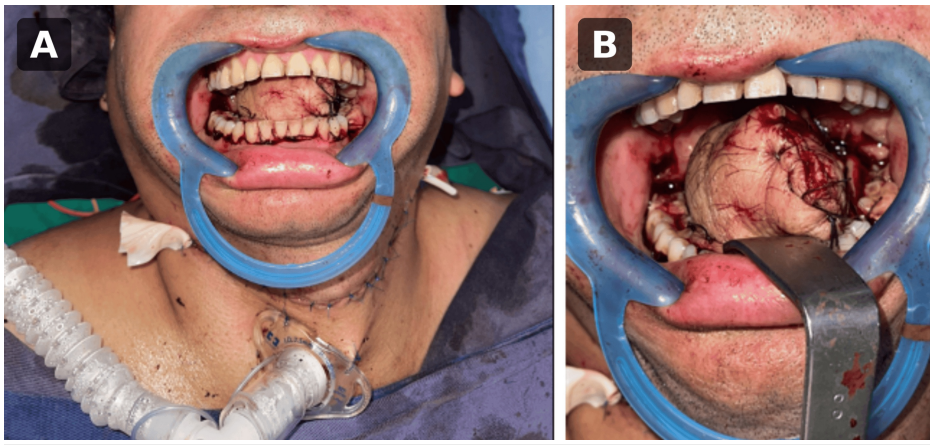


FIGURE 4: Intraoperative images

Intraoperative views after reconstruction of the defect with an anterolateral thigh (ALT) microvascular flap. (A) Neck view with the mouth partially closed, showing the neck dissection wound and the tracheostomy. (B) Oral view with the mouth open.

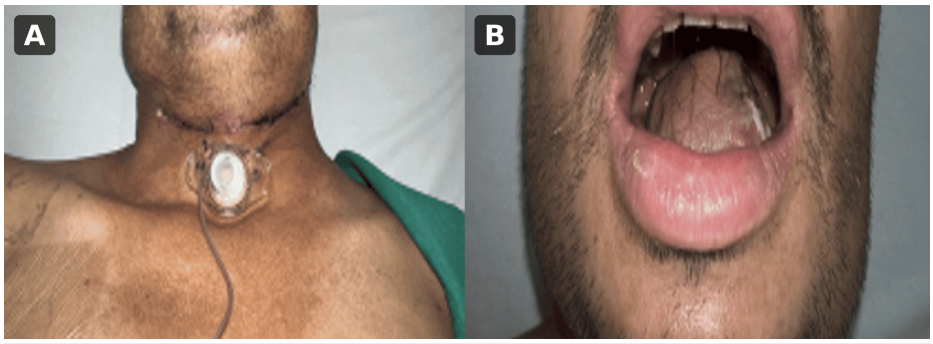


FIGURE 5: Immediate postoperative images

(A, B) Uncomplicated neck surgical wound, functional tracheostomy, and viable microvascular flap.

Definitive histopathological examination of the surgical specimen confirmed an infiltrating, keratinizing, moderately differentiated, ulcerated large-cell squamous cell carcinoma, with focal involvement of the dorsal surface of the tongue tip. The tumor measured 6 × 4.5 cm, with a maximum thickness of 41 mm, and both perineural and lymphovascular invasion were present. Immunohistochemistry demonstrated expression of CK5/6, p63, p40, and p53, absence of p16 expression, and a Ki-67 proliferative index of 70%. The peripheral soft-tissue margins of the anterior tongue, tongue base, and posterior margin were free of malignancy, at a distance greater than 10 mm. Bilateral neck dissection showed no metastatic involvement in any of the 91 examined lymph nodes: 0/2 at level IA, 0/35 in the right neck dissection, and 0/54 in the left. Based on these findings, the definitive pathological stage was pT4aN0M0, stage IVA [7].

Because of the adverse histopathological features identified (pT4a stage, perineural invasion, and lymphovascular invasion), adjuvant image-guided external radiotherapy (IGRT) was administered in 2-Gy fractions to a cumulative dose of 66 Gy (33 fractions) to the surgical bed of the primary tumor site. Elective cervical nodal irradiation was not performed because of the negative bilateral neck dissection; consequently, the neck was not included in the planned target volume, and the tracheostomy stoma was outside the target volume, although a low, non-calculated scattered dose cannot be excluded. Concomitant chemotherapy was likewise not indicated, given the absence of positive margins or extranodal extension.

Adjuvant radiotherapy began nine weeks after surgery, delayed by administrative authorization procedures, and was delivered over approximately 7.5 weeks, with a brief interruption during the year-end holidays. At a follow-up visit during the sixth week of treatment, approximately 15 weeks after surgery, inflammatory

changes and slight skin thickening were noted in the anterior cervical region, without exophytic or vegetant lesions; these were attributed to probable radiodermatitis due to scattered dose, and treatment was continued. At the next visit, nine days later, approximately 16 weeks after surgery and during the last week of treatment, a vegetant, irregular, foul-smelling lesion with rapidly progressive growth and bleeding was identified around the tracheostomy stoma (Figure 6). The patient was referred for expedited evaluation, and contrast-enhanced neck computed tomography demonstrated soft tissue involvement around the stoma, with extension toward the tracheal rings and partial reduction of the tracheal lumen (Figure 7). After assessment by the head and neck surgery team, a biopsy of the lesion was obtained the following day. The patient remained clinically stable and completed the planned radiotherapy. Subsequently, histopathological examination demonstrated poorly differentiated, focally keratinizing squamous cell carcinoma, confirming tumor recurrence at the tracheostomy stoma.

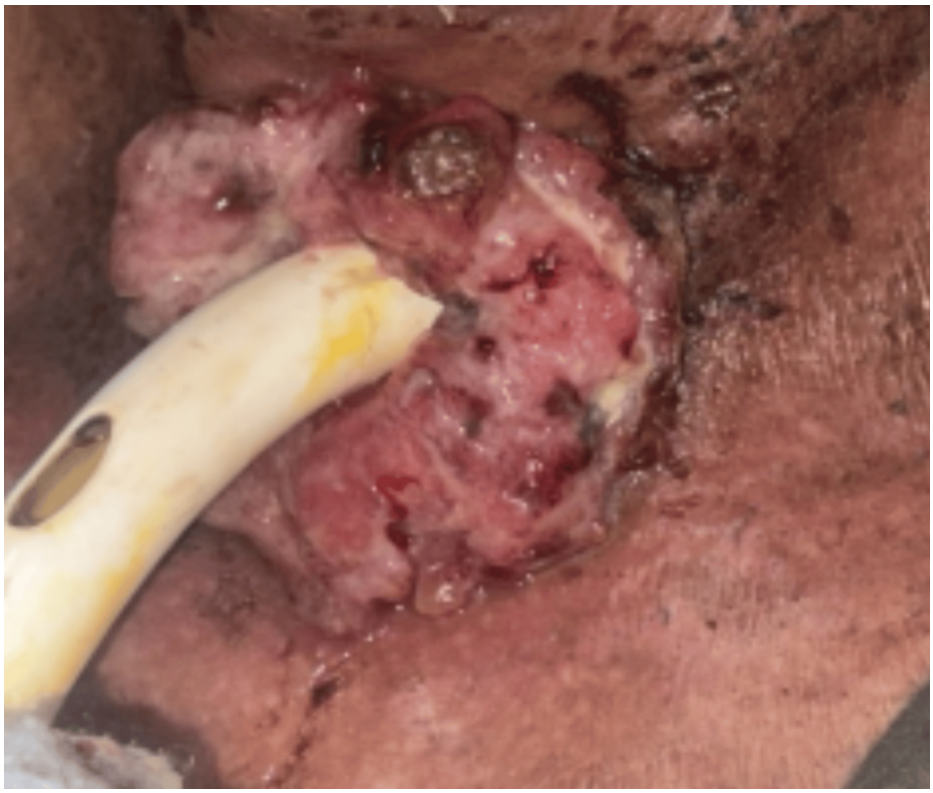


FIGURE 6: Tracheostoma photograph

Vegetant, irregular lesion surrounding the tracheostomy stoma, with the tracheostomy cannula in place.

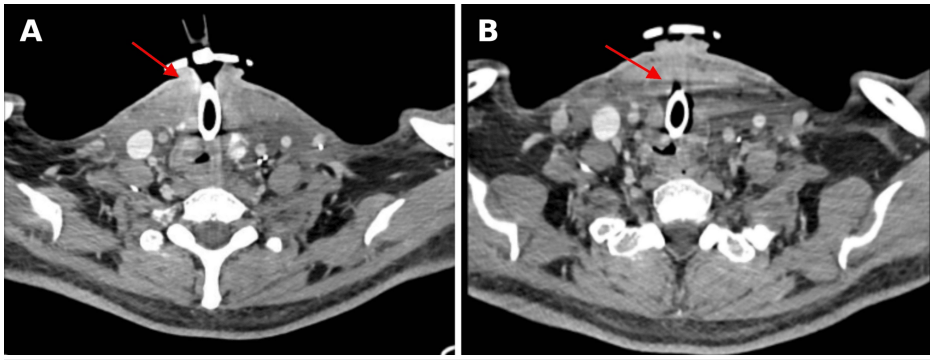


FIGURE 7: Axial contrast-enhanced neck computed tomography at the level of the tracheal stoma

(A, B) Red arrows indicate the exophytic peristomal lesion at the tracheostomy site, with soft-tissue involvement extending toward the tracheal rings and partial narrowing of the tracheal lumen.

With the biopsy result and imaging findings, the case was reassessed by a multidisciplinary head and neck group. Given the extent of the disease and involvement of adjacent structures, the recurrence was considered not amenable to surgical salvage. The radiation oncology team proposed a new course of irradiation directed at the involved region, considering that this cervical area had not been included in the target volume of the previous radiotherapy, while medical oncology proposed cisplatin-based systemic treatment.

Before this new treatment regimen could be initiated, the patient developed acute clinical deterioration requiring hospitalization, characterized by persistent cough, dyspnea, facial edema, and bleeding through the tracheal stoma. Chest computed tomography revealed pulmonary parenchymal abnormalities suggestive of an aspirative and/or infectious process. Laboratory studies showed leukocytosis with neutrophilia, reactive thrombocytosis, and elevated acute-phase reactants, findings clinically compatible with tracheitis and aspiration pneumonia superimposed on locoregional tumor progression.

Despite empiric antibiotic treatment, respiratory symptoms persisted, and rapid clinical and functional deterioration occurred. The patient reached an ECOG performance status of 4, a Karnofsky Performance Status of 20%, a Palliative Performance Scale (PPS) score of 20%, and a Palliative Prognostic Index (PPI) score of 10, indicative of advanced oncologic disease and clinical deterioration with low reversibility.

In conjunction with the pain medicine and palliative care team, a goals-of-care approach focused on symptom control and comfort measures was adopted, with de-escalation of therapeutic interventions. The patient subsequently progressed to the end-of-life phase and died.

Discussion

Tumor recurrence at the site of a temporary tracheostomy performed during the surgical treatment of head and neck squamous cell carcinoma is an infrequent phenomenon that has been documented for several decades. Armstrong and Price described a patient with tongue squamous cell carcinoma who developed an aggressive recurrence at the tracheostomy scar seven months after hemiglossectomy and adjuvant radiotherapy. The disease showed subglottic progression and subsequently required total laryngectomy, with an ultimately fatal outcome [8].

One of the mechanisms proposed to explain this phenomenon is mechanical implantation of tumor cells during the surgical procedure. Atiyah et al. experimentally evaluated this hypothesis by collecting blood from the surgical field during oncologic head and neck resections. Using the Hamburger and Salmon tumor stem cell assay, they identified viable colony-forming tumor cells in one of six evaluable preparations [3]. These findings provide biological plausibility for the possibility that neoplastic cells released during tumor manipulation may contaminate other surgical sites. However, the demonstration of viable tumor cells in the operative field does not by itself establish that direct implantation is the mechanism responsible for stomal recurrence.

In this context, it has been proposed that the sequence of tracheostomy relative to tumor resection may play a relevant role. During procedures that combine tracheostomy, resection of the primary tumor, and neck dissection, a stoma created before completing the resection could be exposed to blood or secretions potentially contaminated with tumor cells. This hypothesis has been considered particularly relevant when tracheostomy is performed before oncologic resection [3,4].

Campbell et al. described two cases of tumor implantation at the tracheostomy site in patients with squamous cell carcinoma of the upper aerodigestive tract [5]. The authors noted that although postoperative radiotherapy included the primary tumor site and the cervical regions considered at risk, the tracheostomy site was left outside the irradiated field. These observations raised the possibility that tumor cells implanted at the stoma could persist in a region not included in the treatment volumes. Based on these cases, the authors highlighted the importance of considering the tracheostomy site during postoperative radiotherapy planning [5]. However, since this evidence was derived from case reports, a causal relationship between the exclusion of the stoma from the irradiated volume and the subsequent development of recurrence cannot be established.

Halfpenny and McGurk subsequently provided observational evidence by analyzing 265 patients who underwent temporary tracheostomy during treatment for head and neck malignancies [4]. Three patients developed stomal recurrence, corresponding to an approximate incidence of 1%. All three cases occurred in the group in which tracheostomy was performed before tumor resection (three of 83 patients), and all had node-positive disease. No stomal recurrences were identified among the 182 patients in whom tracheostomy was performed after resection. Although the small number of events limits the ability to establish definitive associations, these findings suggest that tumor implantation may participate in the pathogenesis of this complication and that surgical sequence may be clinically relevant [4].

Subsequently, Esteller et al. analyzed a cohort of 491 patients with head and neck squamous cell carcinoma who underwent temporary tracheostomy, which, in surgically treated patients, was performed before tumor

resection, and documented six recurrences at the stomal scar, for an incidence of 1.2% [6]. In addition to confirming the low frequency of this phenomenon, the authors observed an unfavorable prognosis after its onset: five of the six patients died within a year, and only one of the three patients who underwent salvage treatment achieved disease control. These results reinforce the clinical relevance of stomal recurrence despite its low incidence.

Hintze et al. described a fungating tumor recurrence at the site of a prior tracheostomy six months after resection of an oral cavity squamous cell carcinoma. This report confirms that this phenomenon continues to be observed in contemporary practice. Notably, the patient had received adjuvant radiotherapy covering the tracheostomy site, indicating that recurrence can occur despite irradiation of the stoma. The authors noted that several mechanisms have been proposed to explain stomal recurrence, but its pathogenesis remains uncertain [9]. Therefore, although mechanical implantation constitutes a potential mechanism to explain this phenomenon, the available evidence does not allow other routes of dissemination or locoregional progression to be ruled out.

The present case shares several elements with those described previously, but also presents features that are particularly relevant. Tracheostomy was performed before tumor resection, during the same procedure in which bilateral compartmental glossectomy and neck dissection were carried out. This surgical sequence was also observed by Halfpenny and McGurk in patients who subsequently developed stomal recurrence [4], raising the possibility that the stoma was exposed to tumor cells during manipulation and resection of the primary tumor.

However, our case shows an important difference from that series. The three patients with stomal recurrence described by Halfpenny and McGurk had node-positive disease, whereas our patient underwent bilateral neck dissection with no evidence of metastasis in any of the 91 lymph nodes examined. Therefore, stomal recurrence occurred in the absence of histopathologically demonstrable nodal disease. This finding does not demonstrate a direct implantation mechanism, but it makes it particularly relevant to consider mechanisms other than conventional cervical nodal progression.

Another noteworthy aspect is the timing of the recurrence. Whereas some previously published cases became apparent several months after surgery [8,9], in our patient, the stomal lesion became clinically evident during the last week of adjuvant radiotherapy, approximately 16 weeks after surgery, which is sooner than in those two reports and was followed by rapid progression. Nine days earlier, inflammatory changes and slight skin thickening had been noted in the anterior cervical region and attributed to radiodermatitis; in retrospect, they may have represented initial manifestations of the recurrence. The lesion showed accelerated vegetant growth, bleeding, and extension toward the tracheal rings, with partial reduction of the tracheal lumen. Histologically, the recurrence corresponded to a poorly differentiated, focally keratinizing squamous cell carcinoma, in contrast with the moderately differentiated primary tumor. This difference may reflect tumor progression with loss of differentiation, or a poorly differentiated component that was not sampled in the primary tumor; interobserver variability in histopathological interpretation may also contribute. In any case, these histological differences do not by themselves allow a specific mechanism of dissemination to be established.

The relationship between the site of recurrence and the radiotherapy volume constitutes another relevant aspect. In our patient, adjuvant radiotherapy was directed at the surgical bed of the primary tumor site and did not include the neck, so the tracheostomy stoma remained outside the target volume, although a low scattered dose cannot be excluded. Campbell et al. had previously noted the possibility of recurrence at the tracheostomy site when it was excluded from the irradiation fields [5]. Because the lesion appeared during radiotherapy, a radiation-induced malignancy, which typically develops years after exposure, appears unlikely. A metachronous second primary tumor cannot be completely excluded in the absence of molecular studies, but the anatomical and temporal context makes recurrence the more plausible explanation.

From the standpoint of adjuvant treatment, the patient had histopathological risk factors that justified postoperative radiotherapy, including a pT4a tumor and the presence of perineural and lymphovascular invasion. In the absence of positive margins or extranodal extension, there was no conventional indication to intensify treatment with concomitant postoperative chemoradiotherapy [10]. Consequently, the occurrence of stomal recurrence should not necessarily be interpreted as a consequence of an inadequate choice of adjuvant modality, but rather as an infrequent pattern of locoregional failure.

Several measures aimed at reducing the potential risk of tumor implantation have been proposed in the literature, including performing tracheostomy after completing tumor resection whenever airway conditions allow, reducing cross-contamination between surgical fields through changes in gloves and instruments, protecting the aerodigestive tract during resection, and irrigating the surgical field before closure [3-6,9]. Likewise, some authors have proposed considering the tracheostomy site during postoperative radiotherapy planning [5], although recurrence has also been reported despite irradiation of the stoma [9]. However, these strategies are supported mainly by biological plausibility, case reports, and retrospective studies with a limited number of events; therefore, their ability to prevent stomal recurrence has not been established through high-quality comparative studies.

This report has the limitations inherent to a single case. Molecular studies were not available, so the clonal relationship between the primary tumor and the stomal lesion could not be assessed, and the contribution of mechanical implantation cannot be established from a single observation.

Taken together, the available evidence indicates that recurrence at the site of a temporary tracheostomy is an exceptional complication, with a reported incidence near 1% and an unfavorable prognosis [4,6]. The present case is particularly relevant because of the rapid onset and progression of a recurrence at the tracheostomy stoma, which became evident approximately 16 weeks after surgery during adjuvant treatment, following oncologic resection with negative margins and bilateral neck dissection with 91 disease-free lymph nodes. Although mechanical implantation of tumor cells constitutes a plausible hypothesis, particularly when the tracheostomy precedes tumor resection, the responsible mechanism cannot be established with certainty. These findings highlight the importance of clinical surveillance of the tracheostomy site and suggest that its anatomic relationship with radiotherapy volumes may merit consideration in selected patients.

Conclusions

Tumor recurrence at the site of a temporary tracheostomy is an infrequent but serious complication of the surgical treatment of head and neck squamous cell carcinoma. Although intraoperative mechanical implantation of tumor cells constitutes a biologically plausible mechanism, a definitive causal relationship could not be established in this case. The present case shows that a recurrence at the tracheostomy stoma can occur in the absence of histopathological nodal disease after extensive bilateral neck dissection, and that it can become evident during adjuvant radiotherapy. In this case, the tracheostomy was performed before tumor resection, and the tumor had a high burden and adverse histopathological features; however, these findings do not allow the mechanism responsible for the recurrence to be determined.

A vegetant, indurated, ulcerated, or hemorrhagic lesion at a tracheostomy stoma in a patient with a history of head and neck squamous cell carcinoma should prompt a targeted oncologic evaluation and, when appropriate, histopathological confirmation. Skin changes near the stoma during radiotherapy, although usually attributable to radiodermatitis, should be monitored closely. Whether considering the tracheostomy site in postoperative radiotherapy planning, or applying specific modifications in surgical technique, can reduce the risk of stomal recurrence remains to be determined, and further studies are needed to establish the true role of tumor implantation in this infrequent but serious complication.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

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Disclosures

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