

CASE REPORT

Tongue metastasis originated from esophageal squamous cell carcinoma: A case report

Seyed Amir Aledavood¹ | Mahsa Akbari Oryani² | Zohreh Pischevar Feizabad³ 

¹Cancer Research Center, Mashhad University of Medical Sciences, Mashhad, Iran

²Department of Pathology, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran

³Student Research Committee, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran

Correspondence

Zohreh Pischevar Feizabad, Cancer Research Center, Omid Hospital, Mashhad University of Medical Sciences, Koohsangi Ave., Shariati Sq., Mashhad, Razavi Khorasan, 9176613775, Iran.
Email: pischevarFZ971@mums.ac.ir

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Abstract

The tongue is a rare site for metastasis. In this report, we present a woman with a history of esophageal squamous cell carcinoma who suffered from two metastatic masses on the left side of her tongue 2 years after completion of definitive chemoradiation. The patient underwent chemotherapy and left hemi-glossectomy.

KEYWORDS

aerodigestive malignancy, esophageal cancer, squamous cell carcinoma, tongue metastasis

1 | INTRODUCTION

The esophageal squamous cell carcinoma is known as locally recurrent disease which tends less to metastasize to distant organs compared with its counterpart, that is, adenocarcinoma.¹ However, distant metastases might happen, mostly involving liver, lung, and bone, compromising the survival of patients, significantly.²

The tongue is a rare site for distant metastasis and only few cases from different origins such as gastrointestinal tract or pulmonary system have been reported.³⁻⁵ Although it is more frequent in patients with disseminated diseases, it could be observed as single site of metastasis or even as the initial presentation of distant metastasis, as well.^{5,6}

In this report, we present a woman with a previous history of esophageal squamous cell carcinoma (ESCC) with

final diagnosis of the metastasis to tongue 2 years after definitive chemoradiation.

2 | CASE PRESENTATION

A 54-year-old woman with a past history of esophageal squamous cell carcinoma was admitted due to two soft tissue masses on the left side of her tongue. The patient did not mention history of smoking, addiction, or using nas-vay. She had no significant past medical history other than her esophageal cancer which was treated by definitive radiation therapy [50 gray/25 fractions during 5 weeks] with concurrent chemotherapy [two cycles of cisplatin (30 mg) and 5-FU (500 mg) day1-3 every 3 weeks]. After treatment, she was followed up every 4 months and had no specific signs and symptoms till 24 months later when

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she complained of persistent lumps on the left side of her tongue which were present since last 4 months.

Physical examination showed there were two separated painless firm masses on the left side of the tongue without any signs of epithelial and mucosal involvement or ulcer. The incisional biopsy showed nondysplastic epithelium lied over a submucosal carcinomatous neoplasm composed of tumoral cell islands with severe atypia and conspicuous nucleoli along with frequent mitosis, necrosis, and lymphovascular invasion (Figure 1A,B). The primary pathologic diagnosis was poorly differentiated carcinoma which subsequent immunohistochemistry evaluation showed positive staining for CK and P63 and negative staining for CK5/6, EMA, and CEA leading to final diagnosis of metastatic high-grade squamous cell carcinoma.

Esophagogastroduodenoscopy and full-body computerized tomography (CT) scans with and without intravenous contrast were done. Barium meal revealed smooth narrowing of middle esophagus. However, the endoscopic biopsy of esophageal stricture was normal.

Cisplatin (cisplatin, 80 mg/m²/day, day 1) plus 5-FU (800 mg/m²/day, days 1–5) chemotherapy regimen was commenced and repeated every 3 weeks. After four cycles of treatment, the lingual lesions have shrunk, significantly, depicted on the physical examination. Subsequently, the patient underwent left hemi-glossectomy, and pathologic examination revealed pseudoepitheliomatous hyperplasia confirming a pathologic complete response (Figure 1C,D). After 1 year of follow-up, she was disease free.

3 | DISCUSSION

The tongue is an unexpected site of distant metastasis for malignant diseases including esophageal cancers and only few reports are available, mostly originated from the esophageal adenocarcinoma.^{5,7} Therefore, the diagnosis of tongue metastasis as a single site of distant metastasis which is originated from esophageal squamous cell carcinoma is a challenging scenario because

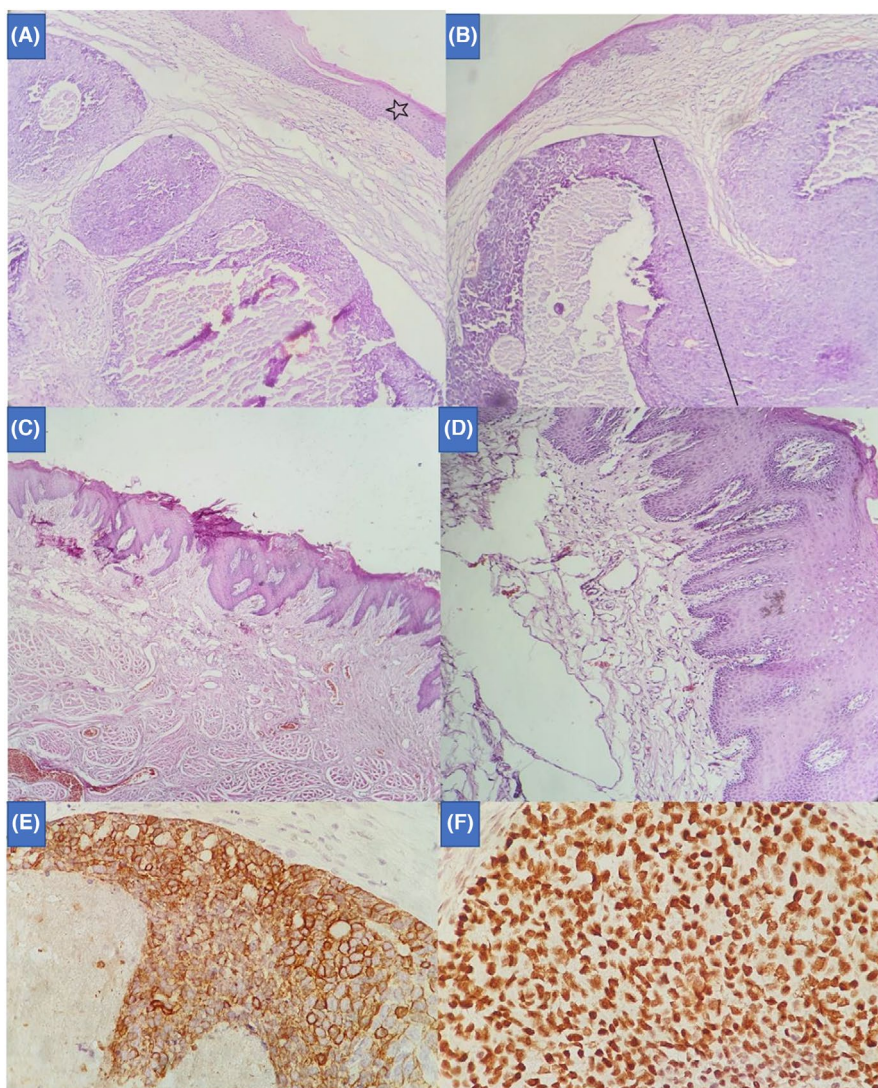


FIGURE 1 A (×10) - B (×10): nondysplastic epithelium (*) lied over a submucosal carcinomatous neoplasm composed of tumoral cell islands with severe atypia (—) and conspicuous nucleoli along with frequent mitosis, necrosis, and lymphovascular invasion. C (×40) - D (×10): Total glossectomy following chemotherapy; achieving pathologic complete response following chemotherapy. E (×40): membranous immunostaining of CK20 was seen in tumoral cells F (×40): A strong nuclear p63 immunostaining was seen in tumoral cells

of its rarity as the location of metastasis and also possible second primary malignancies of the aerodigestive tract due to potential shared predisposing factors and field cancerization.⁸ Histopathologic evaluation might be helpful to distinguish between the primary squamous cell carcinoma of tongue and the metastatic lesions originated from squamous cell carcinoma of other organs. Intact nondysplastic epithelium of tongue is the main characteristic of metastatic lesions as reported in the current case.

Shimizu et al. (2021),⁹ reported an old man with esophageal cancer who had been diagnosed with a metastasis to the tongue during post esophagectomy adjuvant chemotherapy. In contrast to the patient presented by us, the patient reported by Shimizu et al. suffered from disseminated disease with mediastinal lymph nodes metastases and died 1 year after treatment despite the chemotherapy and radiotherapy. Tunio et al. (2014)⁵ reported a case of esophageal adenocarcinoma which had been diagnosed by a tongue metastasis as the initial place of distant metastasis. The patient reported by Tunio et al. died 2 months after surgical resection of tongue mass due to progressive disease, as well.

Patients with metastatic esophageal squamous cell carcinoma usually suffer from disseminated disease and single distant metastasis, especially to the tongue, is extremely uncommon. Despite the positron emission tomography-computed tomography (PET-CT) scan was not performed in our patient, results of the extensive investigations including esophagogastroduodenoscopy and full-body CT scan did not show neither local nor distant recurrent except the lingual lesion.

Because of the rarity of condition, no standard approach for the treatment of patients has been proposed. Considering the morbidity of surgical resection in patients with malignancy of tongue, palliative chemotherapy and or radiotherapy, if there is no overlap with previous radiation plans, are among the best treatment options.^{10,11} The presented patient with solitary distant metastases from esophageal cancer underwent definitive surgery after the preliminary chemotherapy showing a complete response at the pathologic examinations. Definitive surgery can be offered to patients with the isolated recurrence from esophageal cancer without any other evidence of local or distant metastasis and offer a survival benefit for properly selected patients.¹² Beside the conventional chemotherapeutic agent,¹³ innovation of newer treatment approaches such as targeted therapy and immunotherapy,¹⁴⁻¹⁷ introducing newer techniques in delivering the radiotherapy,^{18,19} and finding newer markers to assess the response to treatments^{20,21} may improve the survival of patients with metastatic esophageal cancer including people with distant metastasis to rare sites.

4 | CONCLUSION

The tongue is a rare site for distant metastasis. However, in patients with a history of malignancy, any persistent lingual mass should be assessed carefully, regardless of its appearance, growth rate, ulceration, and neural symptoms.

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We would like to thank the patient for her consent for publication of the data.

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest.

AUTHOR CONTRIBUTIONS

The author listed is the sole author. S.A.A. supervised the case report and contributed to the final version of the manuscript. Z.P.F. contributed to the interpretation of the results and took the lead in writing the manuscript. M.A.O. provided the pathologic images and contributed to the interpretation of them. All authors discussed the results and commented on the manuscript.

ETHICAL APPROVAL

An informed written consent form was obtained from the patient.

CONSENT

An informed written consent form was obtained from the patient.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ORCID

Zohreh Pischevar Feizabad  <https://orcid.org/0000-0001-9527-6226>

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