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Case Report

Adenocarcinoma of the esophagus: a case of solitary metastasis to the tongue

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ABSTRACT

Malignant neoplasms that metastasize to the head and neck remain a rare occurrence. Metastatic lesions to the tongue are even more rarely encountered. The majority of the few reported cases have primary sites in the lung, breast, and kidney. Adenocarcinoma of the esophagus with isolated metastasis to the tongue is a unique occurrence with only a handful of cases with other extensive metastases ever reported. Herein we present the case of a patient with adenocarcinoma of the distal third esophagus who underwent a neoadjuvant therapy followed by margin negative esophageal resection, including negative lymph nodes, who subsequently presented with a solitary distant metastasis to the tongue.

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Introduction

Esophageal cancer remains more common worldwide than in the United States (US), with esophageal cancer approximating 17,290 cancers diagnosed in the US in 2018 [1]. The majority of the cases seen in the US: these include adenocarcinoma (60%) and squamous cell carcinoma (40%) [1]. Both variants have similar 5-year survival rates ranging from 40% for localized tumor to 5% for metastatic cases. Common sites for distant metastasis include lung, lymph nodes, liver, bones, and adrenal glands. Nevertheless, metastasis to uncommon sites have been reported in small series and case reports. The head and neck were found to be the majority of uncommon sites accounting for 42% [4]. Herein, we present a case of adenocarcinoma originating in the distal esophagus with a solitary metastasis to the tongue and discuss management.

Case Report

The patient is a 60-year-old male initially presented to the otolaryngology clinic with a history of left lateral tongue pain. His past medical history is significant for gastroesophageal reflux, hypertension,

diverticulitis, and esophageal adenocarcinoma diagnosed 6 months prior. He denies prior tobacco or alcohol use. He was diagnosed with esophageal adenocarcinoma located in the lower third of the esophagus which was confirmed on computed tomography (CT) scan with 32--42 cm crossing the GE junction (Figure 1), uT3N2 on EUS. He had a positron emission tomography (PET) scan (Figure 2) to complete staging which found no distant metastases. He underwent preoperative radiation therapy- 50.4 Gray in 28 fractions with concurrent carboplatin and taxol (5 cycles). Two months after the completion of chemotherapy and radiation, he underwent a thoracoscopic assisted transhiatal esophagogastrectomy and pyloroplasty; his postoperative course was unremarkable with final pathology showing yT2N0M0 stage 1 disease. He recovered well from these therapies.

On follow up a few months after the esophagogastrectomy he was found to have a 7mm x 7mm, left lateral tongue mass with an overlying clean ulceration that was biopsied revealing normal cellular architecture. The patient then returned to clinic 1 month later with increasing tongue pain and dysphagia. Upon examination, the previously clean ulceration had changed to an exophytic lesion (Figure 3). A biopsy was taken at this

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time with pathology revealing adenocarcinoma consistent with prior primary esophageal adenocarcinoma. He then underwent a repeat PET that revealed an isolated lesion to the left lateral tongue.

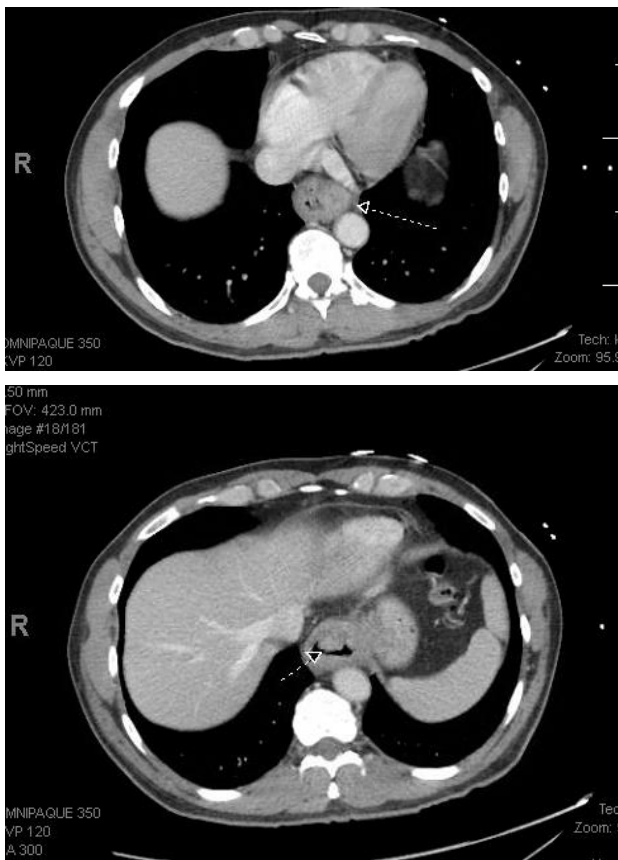


Figure 1: CT scan demonstrating distal esophageal adenocarcinoma.



Figure 2: PET obtained at time of initial diagnosis, demonstrating no distant metastasis, with no abnormal uptake in the tongue.

The patient then underwent a partial glossectomy which revealed tumor with positive inferior margin from original operative specimen. Patient was then taken back for a re-excision and skin graft placement a few days later and adequate negative margins were obtained at that time.

Histopathology from partial glossectomy revealed Invasive adenocarcinoma, moderately differentiated (Figure 4). Tumor morphology was consistent with a metastasis from the known primary esophageal adenocarcinoma with perineural invasion. This was found to also be positive for HER-2 by immunohistochemical staining. This case was then presented at a multi-disciplinary meeting. It was discussed that this rapid recurrence after the esophagectomy, suggests the risk of recurrence in another site is significant. There is limited data regarding adjuvant therapy in this situation, but adjuvant chemotherapy now was selected. Chemotherapy was extrapolated from FOLFOX trials although. After a second medical oncology opinion agreed with proceeding with FOLFOX with the addition of trastuzumab, which the patient is receiving at the time of writing this report [8, 9].



Figure 3: Left lateral tongue exophytic lesion.

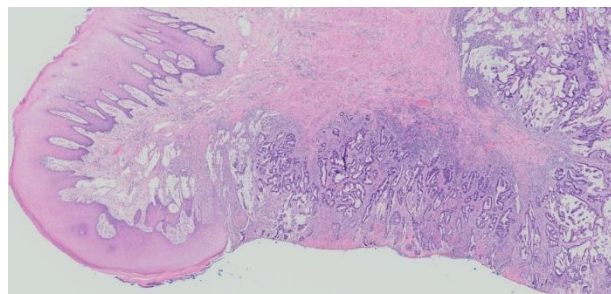


Figure 4: Invasive moderately differentiated adenocarcinoma seen infiltrating the submucosa of the tongue with focal ulceration of the overlying squamous mucosa (Hematoxylin and eosin stain, 40X).

Discussion

Adenocarcinoma of the esophagus has become increasingly prevalent in recent years. Metastatic adenocarcinoma typically presents in lungs, liver, bone and adrenal glands. This case presents with a rare site of solitary metastasis. The lymphatic drainage of the esophagus contains little barrier to spread, and esophageal lymphatics are closely interconnected. Hence, esophagus carcinoma can spread through the entire length of the esophagus via lymphatics and has the potential for anatomic involvement away from the primary lesion. However, we believe this lesion arrived in the tongue via hematogenous metastasis.

Patients having metastatic esophageal cancer are typically found to have multiple sites of disease, representing incurable disease. The goals of therapy include palliation of symptoms as well as prolongation of survival. Patients presenting with a distant metastasis demonstrate a poor 5-year survival rate approximating 5% [1]. However, limited data is available concerning survival rates for isolated metastatic lesions. A case report published in 2002, reported a metastatic lesion to the tongue arising initially from an adenocarcinoma of the distal esophagus. This patient was identified to have subsequent lymph node involvement as well as lung metastasis and died of disease following tongue mass excision [5]. The case in this report the authors believe to be unique in that the tongue was a solitary site of metastasis. Long term survival is possible after resection of solitary metastases from esophageal cancer. There is a study reviewing 30 years of metastatic tumors of the adrenal glands. This study revealed aggressive surgical approach addressing adrenal metastasis can alter long-term survival prognosis including from esophageal cancer [6]. The authors are hopeful, given the early detection of the tongue metastasis and aggressive surgical resection of the isolated lesion will improve the prognosis of this patient. He clearly has decreased symptoms after resection of the tongue metastasis.

In conclusion, adenocarcinoma of the esophagus has a potential for rare, isolated metastasis to distant sites. Review of the literature revealed very limited data regarding management of these lesions. However, aggressive surgical management of the tongue metastasis with adjuvant chemotherapy seems warranted and may improve the prognosis of patients with isolated metastasis.

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