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Esophageal Adenocarcinoma Metastasizing to a Solitary Fibrous Tumor: An Unprecedented Case of Tumor-to-tumor Metastasis

UZAIR JOGIAT[1], CHANTALLE GRANT[1], ERIC L.R. BEDARD[1], KAROLYN AU[1], REMEGIO MAGLANTAY[2] and DAVID WILLIAMS[1]

[1]*Department of Surgery, University of Alberta, Edmonton, AB, Canada;*
[2]*Department of Laboratory Medicine and Pathology, University of Alberta, Edmonton, AB, Canada*

Abstract. *Background: Tumor-to-tumor (TMT) metastasis is a rare phenomenon in which a primary malignancy undergoes metastasis to an additional synchronous or metachronous primary tumor. Case Report: This is a case report of a tumor-to-tumor metastasis from a poorly differentiated adenocarcinoma of the esophagus to a solitary fibrous tumor (SFT) of the right posterior neck, in a 70-year-old-male with a solitary right vertebral artery. After appropriate work-up and involvement of the necessary specialties, the patient underwent a complex surgical resection with negative margins. Conclusion: We present the unique case of a patient with TMT from esophageal adenocarcinoma to an SFT in the posterior neck, not previously reported in the literature. This rare condition with unique oncologic implications highlights the need for a multidisciplinary approach, in this case involving thoracic surgery, head-and-neck surgery, medical oncology, radiation oncology, pathology, and neurosurgical sub-specialty services.*

Tumor-to-tumor (TMT) metastasis is a rare phenomenon in which a primary malignancy undergoes metastasis to an additional synchronous or metachronous primary tumor (1). We present the case of a 70-year-old male with a posterior neck mass and solitary vertebral artery. The final pathology of this mass demonstrated a TMT metastasis from a poorly differentiated adenocarcinoma of the esophagus to a solitary fibrous tumor. The management of this complex scenario required a multidisciplinary approach involving expertise from thoracic surgery, head-and-neck surgery, medical oncology, radiation oncology, pathology, and neurosurgical sub-specialty services.

Correspondence to: Uzair Jogiat, MD, Department of Surgery, 8440 112 St NW, University of Alberta, Edmonton, Alberta T6G 2B7, Canada. E-mail: ujogiat@ualberta.ca

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Case Report

Informed consent was obtained for this case report. The patient is a 70-year-old male with a history of high-grade prostate cancer treated with a robotic radical prostatectomy in 2018. He was investigated for a five-centimeter enlarging, painful, right posterior neck mass in December of 2019, which had been noted five year earlier, but had increased in size and become symptomatic over the last 3 months. He underwent an ultrasound-guided core needle biopsy, showing a spindle cell lesion with hemangiopericytoma-like vascular pattern (+CD34, +STAT6, -AE1/AE3) consistent with an SFT. Due to a pacemaker, magnetic resonance imaging of the lesion was contraindicated, and multidisciplinary tumor rounds recommended local excision. The patient subsequently experienced progressive dysphagia and weight loss. An esophagogastroscopy identified a nodular, friable mass in the distal esophagus, and biopsy confirmed a poorly differentiated esophageal adenocarcinoma with signet ring cell features. Staging revealed no evidence of metastatic disease on computed tomography (CT) chest-abdomen-pelvis or positron emission tomography scan. The patient underwent treatment with curative intent, receiving neoadjuvant chemoradiation (CROSS protocol), followed by an R0 Ivor-Lewis esophagectomy with final pathology showing ypT0N1M0.

The patient was subsequently restaged for his SFT. A CT of the soft tissue in the neck in January 2021, demonstrated interval enlargement with erosion of the right occipital bone and a small amount of intra-cranial extension. A subsequent cerebral angiogram identified vascular branches from the right vertebral artery, supplying the tumor at the level of V3 (Figure 1). In addition, the left vertebral artery terminated in extracranial muscle branches. Since the right vertebral artery filled both posterior inferior cerebellar arteries and the basilar artery, its preservation was critical. The patient underwent a right extended neck dissection with planned resection of involved occipital bone and dura. Intra-operatively, however,

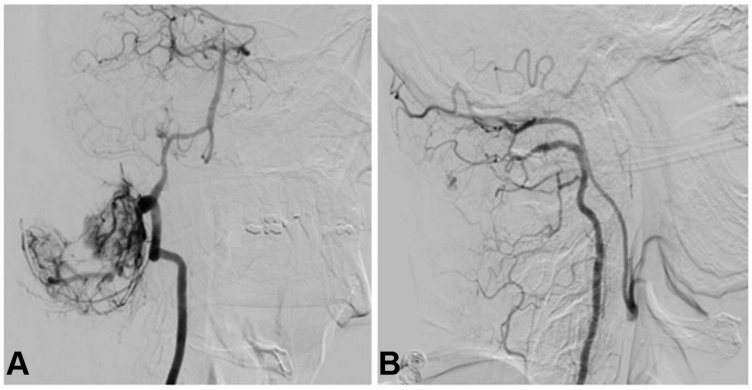

Figure 1. *Cerebral aAngiogram. (A) Right vertebral artery injection, right anterior oblique projection. Arterial supply from branches of the right occipital artery and muscular branches of the V3 segment of the right vertebral artery result in extensive vascular blush in the posterior neck solitary fibrous tumourSFT. The left posterior inferior cerebellar artery and basilar artery fill from the right vertebral artery. (B) Left vertebral artery injection, lateral projection. The left vertebral artery ends in muscular branches with no intracranial communication with the basilar artery.*

the tumor appeared encapsulated with no invasion of bone or dura. The main trunk of the right vertebral artery was not involved in the tumor, allowing for a complete resection of the SFT with preservation of the solitary right vertebral artery. The final pathology demonstrated a margin-negative SFT with a metastatic deposit of poorly differentiated adenocarcinoma (Figure 2). Staining for prostate markers PSA and NKX3.1 were negative, and the immunotype and morphology of the deposit was consistent with esophageal adenocarcinoma (Figure 3).

Discussion

The multidisciplinary management of this patient resulted in successful treatment of both primary malignancies. Prompt identification of the esophageal adenocarcinoma prevented delay in staging and receipt of curative intent therapy, resulting in a favorable oncologic outcome. Re-staging of the SFT enabled identification of tumor erosion into the occipital bone. Cerebral angiogram identified tumor supply from a dominant right vertebral artery, allowing for early consultation of neurosurgery and a combined operation. The involvement of a multidisciplinary tumor group, at the initial diagnosis of the SFT, after re-staging of the SFT, and after resection of the SFT with negative margins, guided clinical management.

Lu *et al*. report a case of esophageal cancer TMT to an intracranial paraganglioma, treated with a primary resection of the paraganglioma and palliative radiotherapy after discovery of the esophageal malignancy (2). Koyama *et al*. report a case of esophageal cancer TMT to gastric cancer, which was treated with primary resection of both lesions (3). The implications of TMT on the staging of the primary tumor, especially within the setting of esophageal cancer – a malignancy known for its invasive nature and poor prognosis, remain unclear. The presence of TMT in the SFT upstages the esophageal adenocarcinoma to oligometastatic disease. In this setting, a personalized approach to management is advocated, with a recent systematic review suggesting a survival benefit for resection of both the primary tumor and the isolated metastasis (4). Upon review at multidisciplinary tumor boards, the decision was made to avoid adjuvant radiation at this time, with ongoing observation to monitor for recurrence.

Conclusion

We present the unique case of a patient with TMT from esophageal adenocarcinoma to a SFT in the posterior neck, not previously reported in the literature. The patient underwent successful management of both malignancies, highlighting the need for early involvement of a multidisciplinary team in the treatment of this rare oncologic phenomenon.

Conflicts of Interest

The Authors have no conflicts of interest to declare in relation to this study.

Authors' Contributions

UJ and CG drafted the manuscript. CG, EB, KA, RM, and DW contributed to patient care. UJ, CG, EB, and DW performed the literature search. UJ, CG, EB, KA, RM, and DW participated in the critical revision of the article. All Authors read and approved the final article for publication.

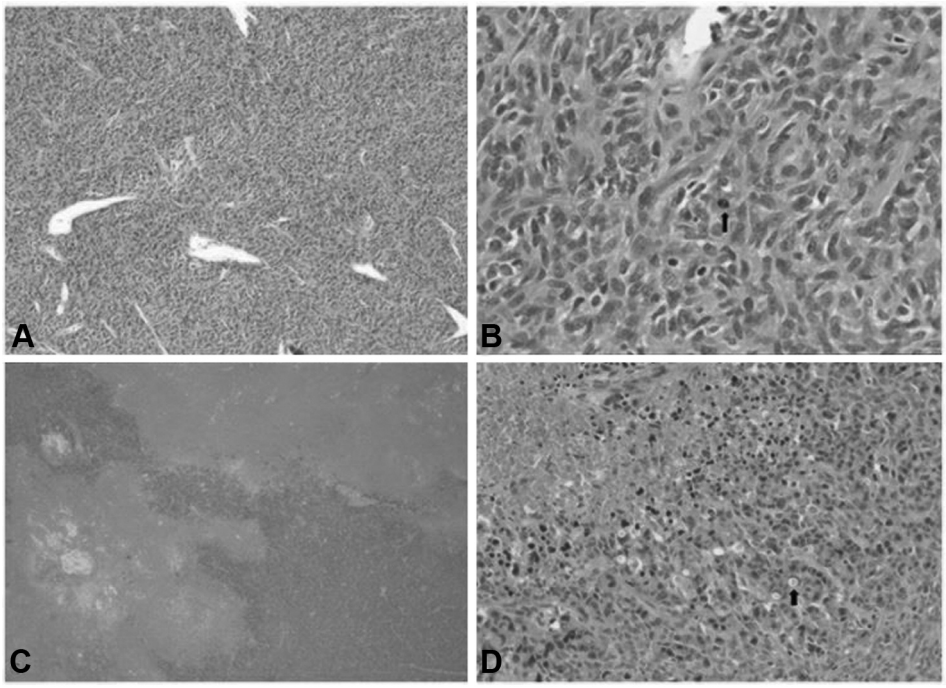

Figure 2. *Histopathology of solitary fibrous tumour (SFT). (A) and (B) show areas of SFT with cellular foci of haphazardly arranged ovoid to fusiform spindle cells, dilated, hyalinized staghorn-like vessels, and focal mitotic activity (arrow). (C) Shows cellular areas corresponding to the SFT (blue) and necrotic foci (pink). (D) High magnification (20×) image of necrotic and viable tumor interface exhibiting highly atypical cells with enlarged hyperchromatic nuclei and abundant eosinophilic cytoplasm with occasional cytoplasmic vacuolization imparting a "signet-ring" appearance (arrow).*

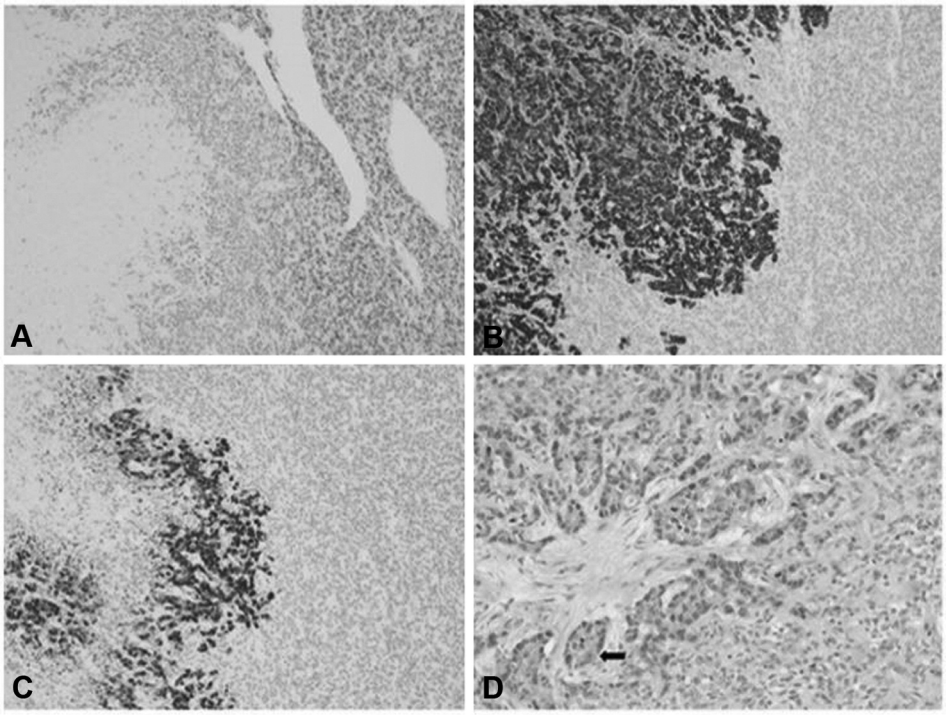

Figure 3. *Special stains applied to histopathology slides of solitary fibrous tumour (SFT). (A) Immunohistochemistry for STAT6 is positive on the SFT component and negative on the poorly differentiated adenocarcinoma. (B and C) AE1/AE3 and CDX2 are positive on the poorly differentiated adenocarcinoma and negative on the SFT. (D) Mucicarmine highlights intracytoplasmic mucin on the adenocarcinoma (arrow).*

References

1 Campbell LV Jr, Gilbert E, Chamberlain CR Jr and Watne AL: Metastases of cancer to cancer. Cancer *22(3)*: 635-643, 1968. PMID: 5673241. DOI: 10.1002/1097-0142(196809)22:3<635::aid-cncr2820220320>3.0.co;2-o

2 Lu JQ, Khalil M, Hu W, Sutherland GR and Clark AW: Tumor-to-tumor metastasis: esophageal carcinoma metastatic to an intracranial paraganglioma. J Neurosurg *110(4)*: 744-748, 2009. PMID: 19072308. DOI: 10.3171/2008.9.JNS08397

3 Koyama T, Matsumori M, Nakamura K and Fujimori T: Metastasis of cancer to cancer: report of a case of esophageal carcinoma metastasizing to early gastric cancer. Jpn J Surg *21(3)*: 352-356, 1991. PMID: 1857041. DOI: 10.1007/BF02470960

4 Jamel S, Tukanova K and Markar S: Detection and management of oligometastatic disease in oesophageal cancer and identification of prognostic factors: A systematic review. World J Gastrointest Oncol *11(9)*: 741-749, 2019. PMID: 31558978. DOI: 10.4251/wjgo.v11.i9.741

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