

Case
Report

Solitary Cardiac Metastasis from Esophageal Cancer

Miho Akabane, MD,¹ Masayuki Urabe, MD, PhD,¹ Yu Ohkura, MD, PhD,^{1,2}
Shusuke Haruta, MD, PhD,¹ Masaki Ueno, MD, PhD,^{1,2} and Harushi Udagawa, MD, PhD^{1,2}

A 72-year-old woman with past medical history of rectal cancer resection (adenocarcinoma, pT3N1aM0) presented with a 2-month history of dysphagia. Imaging studies found a thoracic esophageal cancer, for which subtotal esophagectomy with gastric conduit reconstruction via retrosternal route followed by chemoradiotherapy were performed (squamous cell carcinoma, pT4N1M0, RM1). Seven months after the esophagectomy, a contrast-enhanced computed tomography (CT) demonstrated a new asymptomatic mass inside the right atrium. A thrombus or a tumorous lesion was suspected. Positron emission tomography (PET)/CT showed abnormal uptake in the mass. After a thorough discussion by a multidisciplinary oncology group, we performed 1-week anticoagulant therapy first, resulting in mass enlargement. Then tumorectomy was carried out. The final pathological findings revealed that the mass was squamous cell carcinoma, yielding the diagnosis of cardiac metastasis from esophageal cancer. The patient's postoperative course was unremarkable. PET/CT may help to estimate malignancy and to omit invasive heart surgery.

Keywords: cardiac metastasis, esophageal cancer, resection, positron emission tomography/computed tomography

Introduction

Esophageal cancer is a debilitating disease with a poor prognosis. This disease remains one of the leading causes of cancer-related deaths worldwide. The existence of distant metastases is related to poor outcomes in

patients with esophageal cancer.¹⁾ Nonetheless, surgical resection can improve overall survival in patients with esophageal cancer in the form of oligometastases (i.e., ≤ 5 lesions in a single domain).²⁾ In case of solitary cardiac metastasis, however, the treatment strategy has not yet been established globally because of its rarity. Besides, the therapeutic decision-making remains challenging due to the difficulty of differentiation from a thrombus or a benign tumor, including cardiac myxoma. In this report, we describe a case of solitary cardiac metastasis from esophageal cancer, which was resected after a meticulous assessment, leading to a definitive diagnosis.

Case Report

A 72-year-old Japanese woman with a history of persistent atrial fibrillation was admitted to our institution due to a 2-month history of continuous dysphagia. She

¹Department of Gastroenterological Surgery, Toranomon Hospital, Tokyo, Japan

²Okinaka Memorial Institute for Medical Research, Tokyo, Japan

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Corresponding author: Miho Akabane, MD. Department of Gastroenterological Surgery, Toranomon Hospital, 2-2-2 Toranomon, Minato-ku, Tokyo 105-8470, Japan

Email: akabane.miho@gmail.com



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had a history of rectal cancer treated by laparoscopic low anterior resection (moderately differentiated adenocarcinoma, pStage T3N1aM0 by AJCC-8th) 5 years earlier in our hospital. She also had pulmonary metastases previously for which video-assisted thoracoscopic lung resections had been performed 4 years and 3 years earlier in our institution. Esophagogastroduodenoscopy depicted an ulcerated tumor at the middle third of the esophagus, biopsy from which yielded a pathological diagnosis of squamous cell carcinoma. On endoscopic ultrasound, the tumor depth of invasion was estimated to be T3, and multiple metastases to the cervical and mediastinal lymph node were suspected to be present. No apparent distant metastasis was revealed. After one-course neoadjuvant FP therapy (5-fluorouracil and cisplatin), subtotal esophagectomy with gastric conduit reconstruction via retrosternal route was performed. The excisional wedge was histologically positive because the tumor directly invaded into the left main bronchus, resulting in R2 resection. The final pathological diagnosis was as follows: poorly differentiated squamous cell carcinoma of the esophagus, pT4N1M0 by AJCC-8th. Consequently, postoperative chemoradiotherapy (two courses of FP therapy and 60 Gy totally with daily fractionation of 2 Gy) was conducted.

Seven months after the esophagectomy, a follow-up contrast-enhanced computed tomography (CT) demonstrated a new solitary mass inside the right atrium, despite the patient having no symptomatic complaints (**Fig. 1**). Transthoracic echocardiography depicted a mobile, irregularly shaped multilobular 27 × 15 mm mass attached to right atrial wall. The mass was thought to be an intra-atrial thrombus or a tumorous lesion including metastasis from rectal cancer or esophageal cancer as a differential diagnosis. Although a relatively high plasma D-dimer level (4.1 µg/mL) and past medical history of persistent atrial fibrillation supported the likelihood of a thrombus, positron emission tomography/CT (PET/CT) showed significant accumulation with maximum standardized uptake value 6.0 in the mass, which implied that the mass had malignant viability. The serum tumor markers, carcinoembryonic antigen, carbohydrate antigen 19-9, and squamous cell carcinoma antigen, were within their normal ranges. Catheter-based intra-atrial biopsy was considered to be technically difficult according to a cardiology consultation. Surgical biopsy was thus the only way to obtain pathological specimen of the cardiac mass. However, excisional biopsy with right open thoracotomy seemed to be challenging after

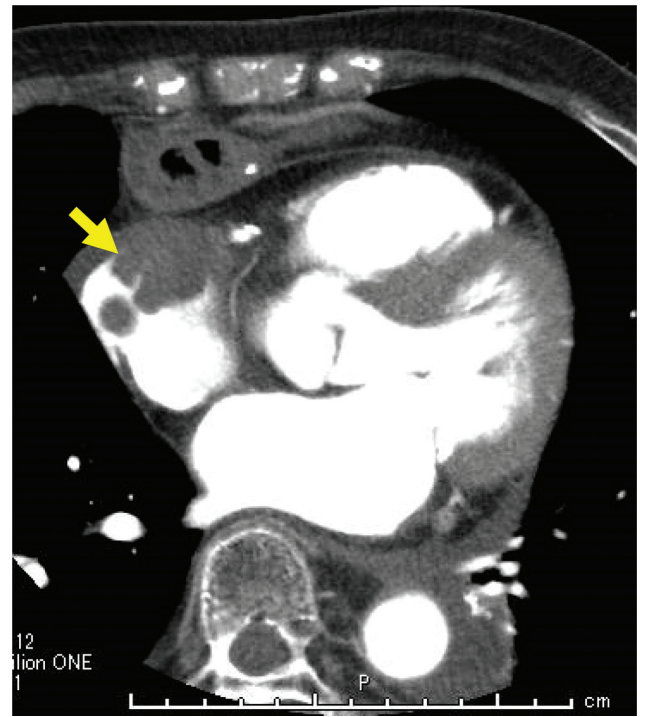


Fig. 1 Contrast-enhanced CT demonstrating an irregularly shaped defect of contrast material inside the right atrium (arrow). CT: computed tomography

retrosternal gastric conduit pull-up. Following an institutional multidisciplinary cancer board discussion, we made a decision to (i) introduce anticoagulant therapy with heparinization and (ii) observe size change of the atrial mass. A week after intravenous heparin administration, contrast-enhanced CT revealed its enlargement to 49 × 16 mm, suggesting the mass not to be a thrombotic lesion but to be a rapid-growing neoplasm. Subsequently, tumorectomy was planned for the suspected malignant tumor to accurately determine diagnosis and to prevent life-threatening morbidities caused by the mass growth, such as congestive heart failure and systemic embolic diseases.

Right lateral thoracotomy was conducted. The solid tumor, occupying the right auricle, was detected under the view of right atrial excision (**Fig. 2A** and **2B**). This tumor was stiff and uneven-shaped, implicating its malignant nature. The mass was resected together with the partial right atrial wall. The defective part of the right atrial wall was repaired with a bovine pericardial patch. Cardiopulmonary bypass time was 48 minutes. The operation time was 148 minutes. Postoperative transthoracic echocardiography showed no residual lesion inside the right atrium.

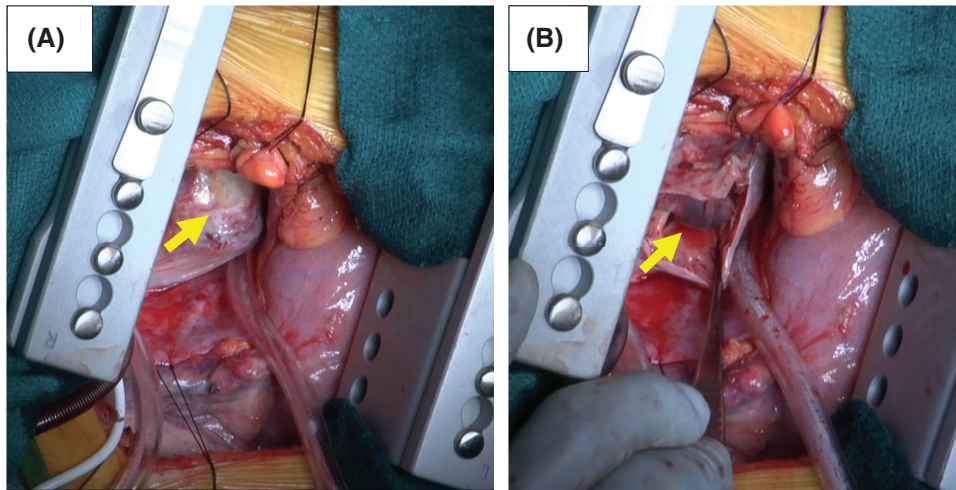


Fig. 2 Intraoperative findings. (A) The right auricle looked stiff due to the existence of the solid tumor (arrow). (B) After the right atrial incision, a floating material protruding from the tumor could be seen (arrow).

The histopathological examination of the resected specimen concluded that the mass was squamous cell carcinoma with partial invasion into the myocardial tissue, yielding the diagnosis of intracardiac metastasis from esophageal cancer (**Fig. 3A** and **3B**).

The postsurgical course was uneventful. To date, 1 month of postoperative outpatient surveillance including contrast-enhanced CT has revealed no evident recurrence of the mass-forming disease. We are planning a regular follow-up imaging study in the outpatient clinic.

Discussion

Solitary cardiac metastasis from esophageal cancer is extremely rare. To the best of our knowledge, only 15 cases of ante-mortem diagnosis have been reported thus far.³⁾ Of them, four cases were simultaneously found at the detection of primary esophageal cancer. In all, 12 cases were solitary cardiac metastases.^{3–17)} Most of the cardiac metastasis are detected on autopsy studies. These studies reported the incidence of cardiac metastasis was 2.3%–18.3% of cancer patients and 0.2%–6.5% of unselected autopsy series.¹⁸⁾ Metastatic diseases are generally formed via the blood stream and the lymphatic system, as well as disseminative spread. Among these, lymphatics is reportedly the predominant route in which esophageal cancer metastasizes to the cardiac wall.¹⁸⁾ Direct invasion is also a major path for reaching the heart. In an anatomical respect, however, the right atrium is unlikely to be subjected to esophageal cancer invasion in contrast to the left atrium. Collectively, intracardiac

metastasis in the present case is most plausibly attributed to lymphatic extension. The possible symptoms of cardiac metastasis are congestive heart failure, syncope, embolism, arrhythmia, electrocardiographic abnormalities, and pericardial effusion. However, these symptoms can abruptly arise, leading to a sudden death, which may suggest the importance of early detection and palliative treatment of cardiac metastasis. According to the past 15 reports of cardiac metastasis from esophageal cancer, the median time from the detection of primary tumor to the onset of cardiac metastasis was 8.9 months.^{3–17)}

The differential diagnosis of cardiac mass is difficult. In case of a cardiac tumor, metastasis is more frequent than a primary tumor. About 75% of primary cardiac tumors are benign and 25% are malignant.¹⁹⁾ Myxoma is the most common benign tumor and angiosarcoma is the most common malignant one, followed by rhabdomyosarcoma.¹⁹⁾ Although cardiac metastasis is usually detected by echocardiography or cardiac magnetic resonance imaging, asymptomatic cardiac metastasis was sometimes detected on PET/CT.²⁰⁾ In the present case, PET/CT was greatly helpful for disclosing malignant potential of the intracardiac mass. It may suggest that PET/CT may serve as an accurate postoperative estimation tool for systemic recurrence.

Decision-making of treatment for a cardiac tumor can also pose a challenge. In general, the therapeutic options are limited due to poor prognosis. Signorelli et al. reported that the median time to death from cardiac metastasis from esophageal cancer was 5.3 months with an overall survival of approximately 18.2 months.³⁾

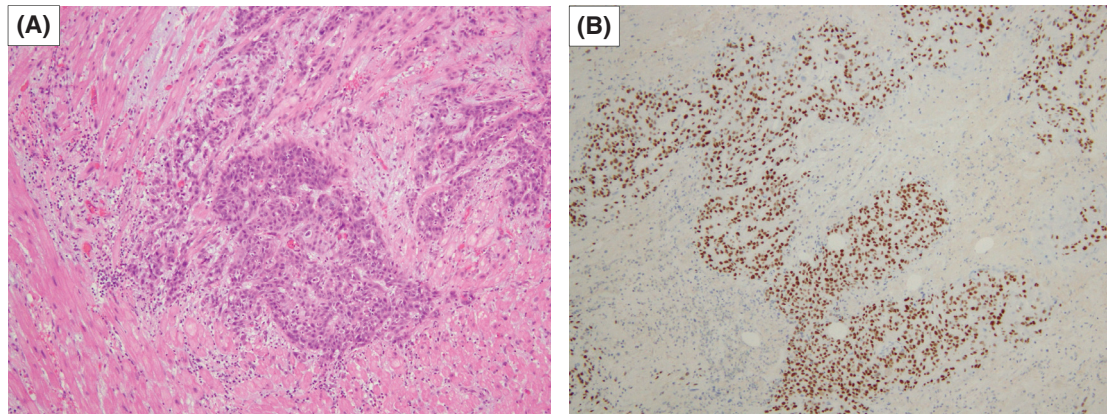


Fig. 3 Pathological findings from the resected specimen. (A) Cancer cells are invading into the heart muscle (Hematoxylin and eosin staining results; $\times 100$). (B) The resected specimen is positive to p40, a marker of squamous cell carcinoma (p40 immunostain; $\times 100$).

Although the treatment of a cardiac tumor provides only palliation, surgical strategy can play an essential part as a diagnostic role because catheter-based atrial biopsy is often technically unfeasible, as in the present case. Of the 15 reports with cardiac metastasis from esophageal cancer, surgical resection was performed in three cases (20.0%).^{3–17} Whether or not surgical removal of cardiac metastasis contributes to prognostic prolongation is totally unknown due to lack of clinical evidence. However, it should not be missed that surgical resection can prevent pulmonary embolism because certain size of mass, whether it is a thrombus or a tumor, can cause pulmonary embolism. Nonetheless, it would be essential to care for the balance between treatment and invasiveness because patients have already undergone highly invasive treatments, namely esophagectomy and chemoradiotherapy.

Besides, no globally approved treatment strategy after resection of cardiac metastasis has been established. Therefore, case-specific discussions have been carried out heretofore. Signorelli et al. insisted that postoperative chemotherapy or radiotherapy should be given to reduce the chance of local recurrence because patients suitable for surgery generally have a better prognosis than inoperable patients.³ In the present case, in-depth discussions by a multidisciplinary oncology group came to two treatment options after resection: chemotherapy (FP therapy) or watching. If the recurrence is identified in case of watching, we will offer a treatment plan according to the location of recurrence. In case of solitary cardiac recurrence, radiotherapy will be offered. In case of sporadic recurrence or the recurrence at the organs where radiotherapy cannot be performed,

chemotherapy will be offered. The patient finally chose watching option after enough consideration.

Conclusion

We encountered an extremely rare case of solitary recurrence to the right atrium from esophageal cancer. Due to difficulty in diagnostic differentiation, therapeutic decision-making for this disease requires careful assessment. PET/CT appears to facilitate reliable judgment of malignancy and might contribute to omit invasive heart surgery.

Informed Consent

Informed consent was obtained from the patient.

Disclosure Statement

All the authors declare that they have no competing interests.

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