Case report

Long-term disease-free survival after resection of recurrent tumor of esophageal cancer with surrounding multiple visceral organs: a case report

Akimori Toyokazu¹, Hiromichi Maeda², Norihiro Kamioka¹, Toshichika Kanagawa¹, Susumu Tsuda¹, Sachi Tsuda¹, Atsunori Takeshita³, Michiya Kobayashi¹, Kazuhiro Hanazaki²

¹ Department of Surgery, Hata-Kenmin Hospital, ² Department of Surgery I, Kochi University, ³ Takeshita Hospital, Kochi, Japan

Abstract

Background: The benefit of resecting recurrent tumor after curative esophagectomy for esophageal cancer remains unclear, especially when it requires resection of multiple visceral organs.

Case presentation: A 56-year-old male patient with previous history of surgical treatment for esophageal achalasia 21 years before was referred to our hospital for treatment of lower thoracic esophageal cancer. He underwent a thoracoscopic esophagectomy and laparoscopic gastric mobilization with curative intent. Nine months after the operation, abdominal computed tomography revealed an intraperitoneal abscess formed along the distal part of the splenic artery. Percutaneous drainage of the abscess and cytological examination diagnosed the tumor as recurrent squamous cell carcinoma from the esophageal cancer. For symptom alleviation and potential cure, the recurrent nodule together with the pancreatic tail, spleen, and left adrenal grand were resected. The pathological examination confirmed recurrent esophageal cancer at the splenic hilar. Three years later, the patient remains disease free.

Conclusion: We experienced a case with metastatic recurrence of lower thoracic esophageal cancer to the hilar of the spleen. When tumor recurrence of esophageal cancer is solitary, an aggressive surgical treatment with multiple-organ resection potentially results in long-term disease-free survival in selected patients.

Key Words: squamous cell carcinoma, multiple resection, spleen

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Introduction

In 2013, there were approximately 442,000 new cases of esophageal cancers and 440,000 deaths due to esophageal cancers reported worldwide¹. Esophageal cancer was therefore declared the ninth-most frequent cancer and the sixth-most common cause of cancer death¹, meaning that esophageal cancer is highly aggressive. Despite efforts to improve patient prognosis with neoadjuvant chemotherapy and surgical resection, esophageal cancer remains frequently associated with tumor recurrence and poor overall survival², ³. The degree of tumor invasion and metastasis to regional lymph nodes is a significant predictor of surgical consequences. When the tumor is pathologically diagnosed as nodal negative, the 5-year survival rate exceeds 75%; however, the survival rate significantly decreases in patients with node-positive esophageal cancer². Tumor recurrence is frequent within the first two years after surgery with the most frequent recurrent site being lymph nodes, followed by hematological metastatic sites such as liver and lung⁴-⁵.

The role of surgical resection for such recurrent tumors is under debate. A recent retrospective study comparing non-resected and resected cases showed favorable overall survival with the surgical treatment⁶, ⁷. Nevertheless, the benefit of multiple organ resection to achieve margin-negative resection of recurrent esophageal cancer remains unclear. In the present report, we describe a patient in which an aggressive surgical treatment with multiple-organ resection for recurrent esophageal cancer resulted in long-term, disease-free survival.

Case presentation

The patient was a 56-year-old male presenting with weight loss and dysphagia. As a 35-year-old (21 years before his current presentation), he underwent surgical treatment for esophageal achalasia, although the details for this were unavailable. Upper gastrointestinal endoscopy revealed a type-2 tumor at the lower thoracic esophagus with a maximum diameter of 5 cm. Pathological examination of a biopsy specimen diagnosed squamous cell carcinoma of the esophagus. Subsequent abdominal
computed tomography (CT) revealed a slightly swollen lymph node around the tumor without any sign of synchronous distant metastasis.

The patient underwent thoracoscopic esophagectomy in the prone position, and laparoscopic gastric mobilization with esophagogastric anastomosis. Postoperative pathological examination diagnosed the tumor as well-differentiated squamous cell carcinoma with invasion outside of the muscularis propria and regional lymph node metastasis around the left gastric artery. The postoperative course was uneventful and he was discharged 22 days after surgery.

Ninth months after the esophagectomy, the patient represented at hospital due to continuous pain at his left upper abdomen. Blood tests revealed an increased white blood cell count and elevated C-reactive protein. CT revealed a space-occupying lesion along the splenic artery from the splenic hilar to the pancreatic tail (Fig. 1). Percutaneous drainage of the abscess was performed and cytological examination revealed recurrence of squamous cell carcinoma. Because of the abscess formation and the lack of evidence for tumor recurrence outside the splenic hilar, the nodule was resected by laparotomy, along with surrounding organs including the spleen, pancreatic tail, and the left adrenal gland (Fig. 2). The postoperative pathological examination confirmed the recurrence of squamous cell carcinoma.

**Fig. 1** Abdominal computed tomography
A mass was identified on the ventral side of the spleen (A). The outer layer of the mass was enhanced with contrast media, while the inside of the mass showed no enhancement, which is suggestive of abscess formation. The mass was compressing the pancreatic tail, suggesting that the resection of pancreatic tail is mandatory for margin-negative tumor resections (B). However, a layer of fat was observed between the left renal vein and the mass.

**Fig. 2** Resected tumor after formalin fixation
The invasion of the tumor to the pancreas and the spleen was evident on macroscopic observation.
esophageal cancer and indicated that the abscess was probably due to ischemia of the spleen from arterial obstruction. Postoperatively, the patient has remained free of recurrence for more than three years.

Discussion

Recent reports indicate the benefit of surgically resecting recurrent tumors of esophageal cancer. A retrospective study reviewing 113 patients with recurrent esophageal cancer demonstrated that the median survival length after recurrence was 92.1 months when surgical resection was performed, while the prognosis without surgical resection, but with chemo/radiation therapy, was comparably poor. The findings also indicated that resection of pulmonary metastases in addition to localized node metastasis could lead to favorable results.

Despite these recent studies, the indication for surgical treatment of recurrent tumors necessitating multiple organ resection remains vague and must be determined on a patient-to-patient basis. In general, the treatment of tumors invading to the adjacent organs would be chemoradiation, especially when the tumor is asymptomatic. In the present case, however, the nodule at the ventral side of the spleen was considered to be responsible for splenic abscess, and the effects of drainage and antibiotics administration was judged to be limited. Therefore, a multiple organ resection to achieve complete removal of the tumor and abscess was planned. As the result, our patient has remained without chemotherapy and recurrence for more than three years. Although this is a single experience in a selected patient, our case may suggest that aggressive surgical resection of a solitary recurrent esophageal cancer has a significant role in treating similar cases.

In the present patient, the route of metastasis remains unclear, although dissemination of the primary esophageal tumor was unlikely because the resected margin at the esophagectomy was negative. Hematogenic metastasis was also unlikely because splenic hilar is quite uncommon site for metastasis. Although the pathological examination revealed no lymph node structures around the metastatic tumor, we consider that metastasis to the splenic hilar lymph node was most likely in this case. Generally, lymph node metastasis at the splenic hilar is scarcely observed in esophageal cancer, and in this case we propose that such a metastatic route was facilitated by destruction of the lymphatic drainage system after the surgery for esophageal achalasia. Following on from that assumption, we recommend that this regional lymph node should be closely observed post-surgery when resection is performed around the esophagus for benign esophageal disease such as achalasia or gastroesophageal hernia. Further accumulation of similar cases is necessary to prove this hypothesis.

In conclusion, this case demonstrated that aggressive surgical resection of solitary recurrent tumor involving multiple organs could lead to long-term, disease-free survival in selected patients. We consider that a precise guide for aggressive surgical resection for recurrence after curative esophagectomy for esophageal cancer should be established.

Reference

1) Global Burden of Disease Cancer Collaboration, Fitzmaurice C, Dicker D, Pain A, Hanavi H, Moradi-Lakeh M, Machtyre MF, Allen C, Hansen G, Woodbrook R, Wolfe C, et al. The Global Burden of Cancer 2013. JAMA Oncol. 2015; 1: 905-27.
2) Yamasaki M, Miyata H, Miyazaki Y, Takahashi T, Kurokawa Y, Nakajima K, Takiguchi S, Mori M, Doki Y. Evaluation of the nodal status in the 7th edition of the UICC-TNM classification for esophageal squamous cell carcinoma: proposed modifications for improved survival stratification: impact of lymph node metastases on overall survival after esophagectomy. Ann Surg Oncol. 2014; 21: 2850-6.
3) Yoshida N, Baba Y, Shiigaki H, Harada K, Iwatsuki M, Sakamoto Y, Miyamoto H, Kurashige J, Kosumi K, Tokunaga R, Watanabe M, Baba H. Risk factors of early recurrence within 6 months after esophagectomy following neoadjuvant chemotherapy for resectable advanced esophagealsquamous cell carcinoma. Int J Clin Oncol. 2016 Jun 2. [Epub ahead of print]
4) Nakagawa S, Kanda T, Konogi S, Ohashi M, Suzuki T. Recurrence pattern of squamous cell carcinoma of the thoracic esophagus after extended radical esophagectomy with three-field lymphadenectomy. J Am Coll Surg. 2004; 198: 205-11.
5) Sugiyama M, Morita M, Yoshida R, Ando K, Egashira A, Takefumi O, Saeki H, Oki E, Kakeji Y, Sakaguchi Y, Maehara Y. Patterns and time of recurrence after complete resection of esophageal cancer. Surg Today. 2012; 42: 752-8.
6) Hiyoshi Y, Morita M, Kawano H, Otsu H, Ando K, Ito S, Miyamoto Y, Sakamoto Y, Saeki H, Oki E, Ikeda T, Baba H, Maehara Y. Clinical significance of surgical resection for the recurrence of esophageal cancer after radical esophagectomy. Ann Surg Oncol. 2015; 22: 240-6.
7) Makino T, Yamasaki M, Miyata H, Tanaka K, Takahashi T, Kurokawa Y, Nakajima K, Takiguchi S, Mori M, Doki Y. Solitary Lymph Node Recurrence of Esophageal Squamous Cell Carcinoma: Surgical Failure or Systemic Disease? Ann Surg Oncol. 2016; 23: 2087-93.
8) Kato H, Tachimori Y, Watanabe H, Iizuka T. Analysis on the lymph node metastasis and prognosis of the esophageal carcinoma. Nihon Shokaki Geka Gakkai Zasshi. 1989; 22: 1729-37. [article in Japanese]