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Prolonged survival of a patient with aortogastric fistula treated with combined surgery and endovascular stent placement: A case report

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ABSTRACT

INTRODUCTION: Aortogastric tube fistula is a rare and fatal complication of esophagectomy. The treatment for aortogastric tube fistula with active infection is challenging, wherein a contamination around the fistula can cause a high risk of aneurysm and recurrence of bleeding, even if large amount of bleeding is controlled immediately.

PRESENTATION OF CASE: We present a case of a 54-year-old male patient who underwent lower esophagectomy for esophageal squamous cell carcinoma 22 years ago. He developed aortogastric tube fistula on postoperative day 46. The patient underwent two surgeries and stenting for aortogastric tube fistula and pseudoaneurysm between days 46 and 120 following the first surgery, and digestive reconstruction was performed 6 months after the first surgery. Computed tomography and esophagogastroduodenoscopy were performed periodically, and the postoperative course was uneventful for 22 years. However, the patient died from pneumonia at the age of 76 years. Autopsy findings revealed no recurrence of esophageal cancer, anastomotic complications, or stent issues. The fistula between the aorta and gastric tube was closed with a stent and connective tissue. Intrathoracic findings revealed that the cause of death was severe bilateral pneumonia.

DISCUSSION: Immediate hemodynamics stabilization and interval infection control enabled successful disease management.

CONCLUSION: Tenting for aneurysm was performed under aseptic conditions, and the patient did not experience recurrence of esophageal cancer and stent issues, which contributed to the long-term survival of 22 years.

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1. Introduction

Aortogastric tube fistula is a rare and fatal complication of esophagectomy. Anastomotic leakage after esophageal reconstruction has been increasingly reported as a significant complication with very high mortality (30 %) [1]. The treatment for aortogastric tube fistula with active infection is challenging, wherein a contamination around the fistula can cause a high risk of aneurysm and recurrence of bleeding even if large amount of bleeding is controlled immediately [1–3]. We herein present the latest status of a patient who was initially reported by Sato et al. in 1999 as the

first surviving case of aortogastric tube fistula after esophagectomy and subsequent endovascular graft placement [2]. The patient had an uneventful course for 22 years after surgery and died from pneumonia. To the best of our knowledge, this is the first report of a patient with long-term survival after three surgeries and stent placement for aortogastric tube fistula in the days without endovascular stent technique. This study has been written in accordance with the SCARE criteria [4].

2. Presentation of case

A 54-year-old male patient with a history of hypertension underwent lower esophagectomy for esophageal squamous cell carcinoma 22 years ago. The surgery was performed by left thoracoabdominal esophagectomy, and gastric tube was reconstructed through the posterior mediastinal route. The pathological diagnosis was T1bN1M0, stage IIB esophageal squamous cell carcinoma according to the 8th edition of UICC staging. Postoperative enhanced computed tomography (CT) scan revealed minor leak-

Abbreviations: CT, computed tomography.

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Table 1
Summary of progress during treatment for aortogastric tube fistula.

Surgery no.	Operation	After first operation	Findings
1	Esophagectomy	–	Left thoracoabdominal esophagectomy, posterior mediastinal route
2	Control the bleeding	46 days	Resection of esophagus and gastric tube and direct suture of the aorta
3	Patch closure	70 days	Resection of pseudoaneurysm and closure using iliac artery patch
4	Stenting	4 months	Modified self-expandable Dacron graft stenting
5	Reconstruction	7 months	Re-reconstruction using jejunum through subcutaneous route

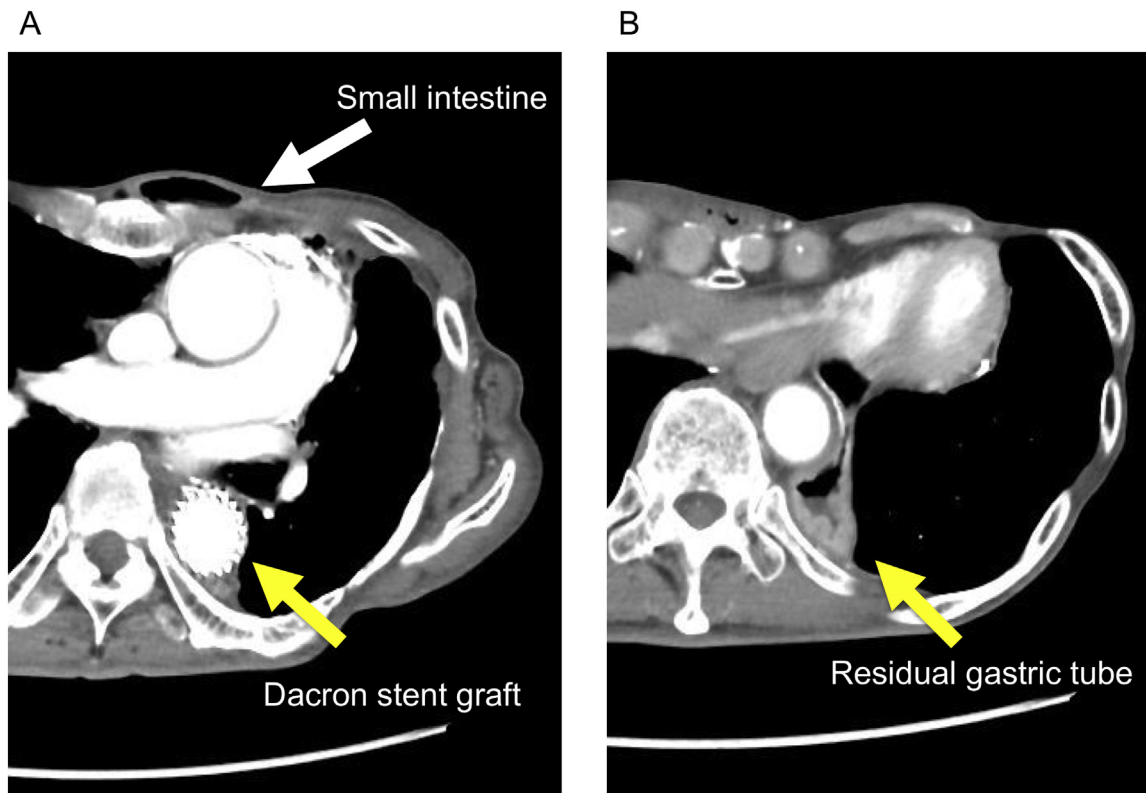


Fig. 1. Representative images of follow-up computed tomography findings. (A) There were no problems with the endovascular stent (Yellow arrow). Roux-en-Y reconstruction with jejunum via the subcutaneous route (White arrow). (B) There was no evidence of local recurrence in the residual gastric tube.

age and a small air space along the anastomotic suture line; thus, the patient was followed with repetitive CT scans and resumption of oral feeding was delayed. However, the patient experienced a large volume of hematemesis and went into shock on postoperative day 46; he was determined to have developed an aortogastric tube fistula. In the second surgery, the esophagus and gastric tube around the anastomosis were partially resected, the perforation site in the aorta was treated quickly by direct closure, and a cervical esophagostomy was constructed. Despite antibiotic treatment, a pseudoaneurysm at the closure suture site developed 19 days after the surgery for aortogastric tube fistula. In the third surgery performed 70 days after the first surgery, the granulation tissue around the pseudoaneurysm was removed and the aorta was closed with an iliac artery patch. Four months after the first surgery, the CT scan and angiography revealed the recurrence of pseudoaneurysm. Results from the bacterial culture with aortic wall in the third operation revealed that the aneurysm was an aseptic condition, and the blood culture was negative after the third operation. Therefore, a modified Dacron graft with a self-expandable stent was placed in the descending aorta. So far, the patient underwent two surgeries and stenting for aortogastric tube fistula and pseudoaneurysm between days 46 and 120 following the first surgery. Roux-en-Y reconstruction using jejunum through the subcuta-

neous route was performed six months after the first surgery (Table 1).

Computed tomography (Fig. 1) and esophagogastroduodenoscopy were performed periodically, and the postoperative course remained uneventful. The patient developed strangulation ileus at the age of 76 years and underwent emergency operation. The patient's condition deteriorated due to overall disuse atrophy, after which he developed repetitive aspiration pneumonia and eventually died as a result of severe pneumonia. Autopsy findings revealed no local recurrence of esophageal cancer, metastasis, or anastomotic or stent issues. The fistula between the aorta and gastric tube was closed with a stent and connective tissue (Fig. 2). Intrathoracic findings revealed that the cause of death was severe bilateral pneumonia.

3. Discussion

This is the first case of a patient with a long-term survival of more than 22 years after treatment for aortogastric tube fistula, a fatal complication of anastomotic leakage, pseudoaneurysm, and ulcer [3]. The present patient was previously reported in 1999 as the first patient surviving surgery for aortogastric tube fistula. Since then, only one of the six reported patients with aortogastric tube

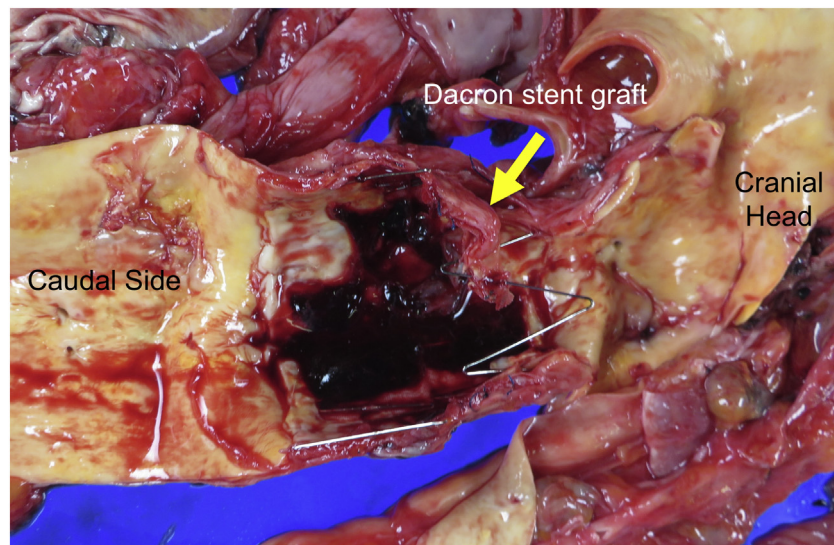


Fig. 2. Representative images of autopsy findings.

The fistula between the aorta and gastric tube was not identified because the fistula was closed by the stent and connective tissue, and there were no problems with the stent (Yellow arrow).

Table 2

Reported cases of aortogastric and aorto-esophageal fistula induced by anastomotic leakage after esophagectomy.

Year, Ref No.	Age	Sex	Surgical procedure	Outcome
2002 [1]	66	M	Endovascular stent	Died (7 months)
2005 [4]	65	M	Direct suture, EVER	Survived (2 years)
2013 [3]	–	–	Patch closure	Died (17 days)
2017 [5]	50	M	TEVER	Survived (1 month)
2017 [5]	69	F	TEVER	Died (88 days)
2017 [5]	62	M	TEVER	Survived (1 month)
Present case	54	M	Surgery and stenting *	Survived (22 years)

Ref No; Reference number.

* Surgery and stenting included direct suture, patch closure, stenting, and reconstruction.

fistula due to anastomotic leakage have survived for two years [1,3,5,6]. The 30-day survival was achieved in four of the patients. All patients underwent endovascular stents placement, and only one patient underwent primary closure before stent placement (Table 2).

Several important features in the present case might have contributed to the successful management of aortogastric tube fistula. The main aim of treatment for aortogastric tube fistula is bleeding control and hemodynamics stabilization [5,7]. Early diagnosis and immediate surgical intervention are necessary for patient survival. In the present case, postoperative CT scan revealed a small air space adjacent to the aorta as well as an anastomotic leakage. This observation led to the immediate diagnosis of aortogastric tube fistula when the patient developed hematemesis and shock. Furthermore, anastomotic leakage-induced aortogastric tube fistula is accompanied with local infection in many cases; therefore, the treatment should include infection control. To avoid prosthetic material placement, closure with direct suture and iliac artery patch were performed in the second and third operation, respectively, in the present case [1,3,5,8]. Although aortic pseudoaneurysm developed after the aortic repair with iliac artery patch, there was sufficient time to control the infected site and the patient eventually underwent endovascular stent placement [2].

Present case had two significant challenges. First, albeit a useful technique for aortic aneurysms, implantation of an endograft with thoracic endovascular aortic repair was not approved for use in humans in 1999 [9]. In the present case, a thin-walled,

30-mm woven Dacron graft was arranged with a self-expandable stent, which allowed the control of aneurysm. Second, the local recurrence could only be assessed by CT imaging; esophagogastroduodenoscopy limited the observation of the end of residual gastric tube because of the Roux-en-Y reconstruction using jejunum. However, the patient showed no recurrence of esophageal cancer on CT scans and during autopsy.

4. Conclusions

Immediate hemodynamics stabilization and interval infection control enabled successful disease management. In addition, stenting for aneurysm was performed under aseptic conditions, and the patient did not experience recurrence of esophageal cancer and stent issues, which contributed to the long-term survival of 22 years.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

This article satisfied the consensus of the National Center for Global Health and Medicine Research Ethics Committee/Institutional Review Board.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

RS and KY wrote the paper. KN, NE, TI, and NK contributed to the paper design and coordination. All authors have read and approved the manuscript.

Registration of research studies

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Guarantor

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