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## Conservative treatment of an aortoesophageal fistula after endovascular stent grafting for a thoracic aortic aneurysm

### Authors' Contribution:

- A** Study Design
- B** Data Collection
- C** Statistical Analysis
- D** Data Interpretation
- E** Manuscript Preparation
- F** Literature Search
- G** Funds Collection

Kazuhiro Kasai<sup>ABCDE</sup>, Akira Ushio<sup>ABC</sup>, Yoko Tamura<sup>B</sup>, Kei Sawara<sup>B</sup>, Yukiho Kasai<sup>B</sup>, Kanta Oikawa<sup>B</sup>, Masaki Endo<sup>D</sup>, Yasuhiro Takikawa<sup>D</sup>, Kazuyuki Suzuki<sup>AD</sup>

Division of Gastroenterology and Hepatology, Department of Internal Medicine, Iwate Medical University, Morioka, Iwate, Japan

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### Summary

#### Background:

Aortoesophageal fistula (AEF) is an uncommon condition that presents a problem in therapy because of the high rate of morbidity and mortality associated with its surgical management and the uniformly fatal outcome of medical treatment. In this article we describe a case of secondary AEF after endoluminal stent grafting of the thoracic aorta, which was observed by only conservative management and followed up for 14 months with no signs of recurrent hemorrhage or chronic mediastinitis.

#### Case Report:

A 54-year old man with hepatocellular carcinoma (HCC) was admitted to our hospital because of tarry stool. He had a history of traumatic aneurysm, and undergone segmental replacement with a stent graft three years ago. After admission, Esophagogastroduodenoscopy and computed tomography identified AEF. He was treated conservatively, because his stage of HCC was advanced. Oral intake was prohibited, and the patient received proton pump inhibitors, intravenous hyperalimentation and antibiotics. Afterwards, no signs of hemorrhage were observed. Although oral intake was resumed after that, another bleeding event or development of mediastinitis was not observed. Subsequently, He was received chemotherapy for advanced HCC, and we observed down-staging of his advanced HCC.

#### Conclusions:

Although we observed 14 months survival in our case under conservative management of secondary AEF, it seems that the treatment of secondary AEF should do the operative management.

#### key words:

**aortoesophageal fistula • hepatocellular carcinoma • conservative treatment • secondary aortoesophageal fistula • stent graft**

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#### Author's address:

Kazuhiro Kasai, Division of Gastroenterology and Hepatology, Department of Internal Medicine, Iwate Medical University, Uchimar 19-1, Morioka, Iwate 020-8505, Japan, e-mail: kaz-k@yc4.so-net.ne.jp

## BACKGROUND

Aorto-esophageal fistula (AEF) is an uncommon condition that presents a problem in therapy because of the high rate of morbidity and mortality associated with its surgical management and the uniformly fatal outcome of medical treatment. Primary AEF has been related to aneurysms of the aorta, foreign bodies in the esophagus, esophageal malignancy, traumatic pseudoaneurysm, and other rare causes [1]. Secondary AEF is rarely reported after endoscopic esophageal procedures, operative procedures and endoluminal stenting [2].

Endoluminal exclusion of abdominal aortic aneurysms has been performed in clinical trials and in clinical practice [3]. More recently endoluminal therapy has been extended to the thoracic aorta. The increasing use and growing expertise in this field stimulated the expansion of indications to a wide spectrum of aortic diseases, although the long-term results and complications of thoracic endografting are still unknown [4]. AEF is one of the potential late complications [5], early diagnosis and prompt surgical management for AEF is crucial for survival [6]. Although conservative management may be chosen for secondary AEF, outcome of conservative management is almost invariably fatal due to recurrent hemorrhage or chronic mediastinitis [5].

In this article we describe a case of secondary AEF after endoluminal stent grafting of the thoracic aorta, which was observed by only conservative management and followed up for 14 months with no signs of recurrent hemorrhage or chronic mediastinitis.

## CASE REPORT

A 54-year old man with hepatocellular carcinoma (HCC) was admitted to our hospital because of tarry stool. He presented with back pain, fever and elevated laboratory markers of systemic inflammation. He had a history of traumatic aneurysm of the descending thoracic aorta three years ago. He had undergone post segmental replacement with a stent graft which was constructed by sewing a Dacron graft to a Gianturco-Rosch Z stent (Figure 1). On admission, there was no sign of active bleeding and he was hemodynamically stable. After admission, Esophagogastroduodenoscopy (EGD) was performed. EGD identified esophageal ulcer with evidence of slight ongoing bleeding at 20 cm beyond the dental arch (Figure 2). A subsequent enhanced computed tomography (CT) showed soft tissue density mass and air bubbles inside which suggested an AEF around descending thoracic stent graft (Figure 3). Surgical repair was not undertaken, because his stage of HCC was advanced, and his long-term prognosis was not expectable. He was treated conservatively. Oral intake was prohibited, and the patient received proton pump inhibitors (PPI), intravenous hyperalimentation and antibiotics (sulbactam/cefoperazone (SBT/CPZ)). SBT/CPZ was administered for 4 weeks. PPI was administered for 2 months. During follow up period, no signs of bleeding and mediastinitis were observed.

Two months later, EGD showed regression of the ulcer without ongoing bleeding (Figure 4). Although oral intake was resumed after that, another bleeding event or development of mediastinitis was not observed. Subsequently, He

was received chemotherapy for advanced HCC. After chemotherapy, we observed downstaging of his advanced HCC. The subsequent post-chemotherapy course was uneventful and patient was discharged in good condition at 4 months after admission. At 10 months after discharge from our hospital, he was in good condition without evidence of bleeding and infection.

## DISCUSSION

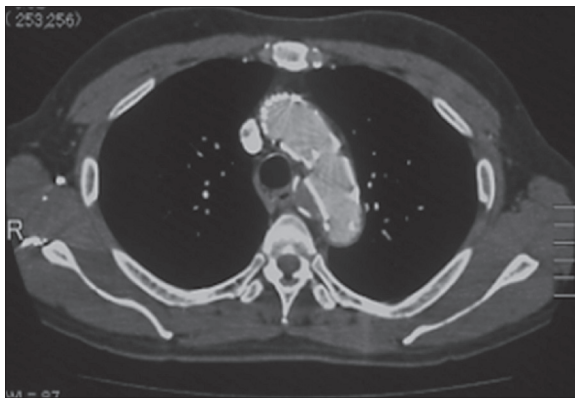
We reported a case with advanced HCC conservatively treated for AEF secondary to endovascular stent-graft repair of descending thoracic aorta.

Aortoenteric fistula following endovascular exclusion of abdominal aortic aneurysm has not been widely reported [7–11]. Moreover, only a few cases of AEF secondary to endovascular stent-graft repair of the thoracic aorta have recently been described [2,5]. One case of such cases have undertaken open surgical repair, and recovered [2]. Although other cases have untreated or treated only medically for secondary AEF, all cases died because of hemorrhagic shock or mediastinitis [5].

The clinical symptom of AEF is a triad of midthoracic pain or dysphagia, a short symptom-free interval, followed by a "herald" hemorrhage and fatal hematemesis. Furthermore, fever and/or sepsis also were described common findings of AEF [12]. Although our case was not observed fatal hematemesis, back pain, fever and findings of systemic inflammation were observed. We guessed the existence of AEF in consideration of these symptoms and a history of traumatic aneurysm of the descending thoracic aorta post segmental replacement with a stent graft.

According to recent reports, it is said that AEF following endovascular repair is absorbed by the following mechanisms; chronic endoleak leading to erosion into the adjacent esophagus, penetration of the stent graft through the aortic wall into the esophagus [5], and ischemic necrosis of the esophageal wall caused by the occlusion of the esophageal arteries that arise directly from the thoracic aorta [13]. Although an EGD for our patient before stent graft implantation showed a normal mucosa of the esophagus and stomach, it developed esophageal ulcer 3 years after stent repair for traumatic aneurysm of the descending thoracic aorta. Considering these facts, ischemic necrosis of the esophageal wall was the most likely mechanism of AEF in our case. The level of the esophageal necrosis corresponded to the segment of the esophagus vascularized by arteries originating directly from the thoracic aorta, under the left bronchus. Although this segment is more susceptible to ischemia because of the lack of collaterals, esophageal ulcer in our case has been improved by use of proton pump inhibitors, intravenous hyperalimentation and antibiotics. Therefore, other mechanisms might exist in our case.

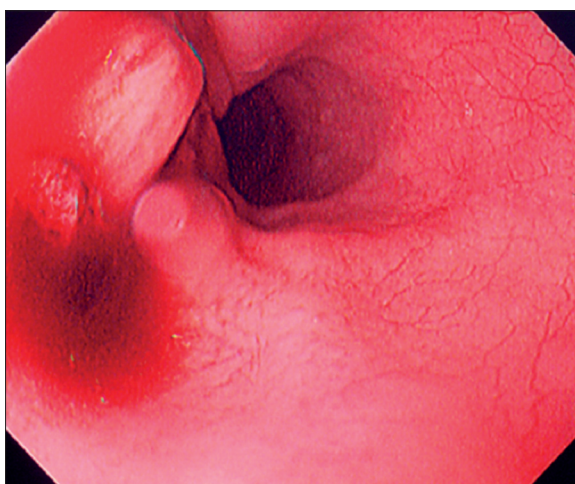
Multiple combinations of treatment options have been used to deal with AEF, including *in situ* arterial reconstruction [1], extra-anatomic bypass with concomitant primary esophageal repair [1,2], or esophagectomy with cervical esophagostomy and secondary restoration of gastrointestinal tract continuity [14]. There is no reasonable alternative for surgery and all surgical candidates should undergo prompt



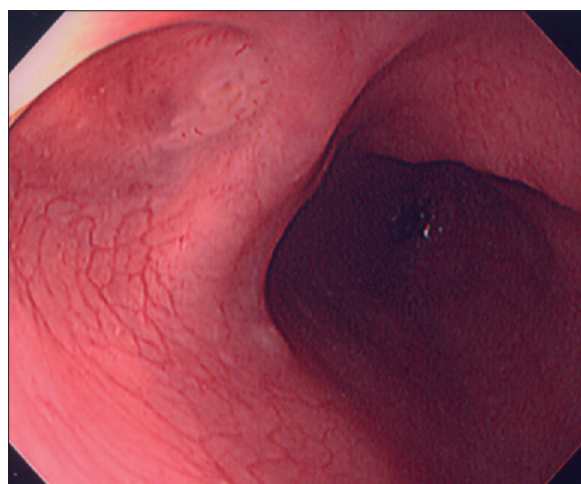
**Figure 1.** Computed tomography done 1 month after endovascular stent grafting. There was no endoleak.



**Figure 3.** Computed tomography demonstrated soft tissue density mass and air bubbles (black arrow) inside which suggested an AEF around descending thoracic stent graft



**Figure 2.** Esophagogastroduodenoscopy demonstrated esophageal ulcer with evidence of slight ongoing bleeding at 20 cm beyond the dental arch.



**Figure 4.** Esophagogastroduodenoscopy demonstrated regression of the ulcer without ongoing bleeding at 20 cm beyond the dental arch.

intervention. However, operative management of AEF has a significant mortality and is frequently complicated by mediastinitis, sepsis, and hemorrhage [15]. We chose conservative management in our case, because his stage of HCC was advanced, and his long-term prognosis was not expectable. Conservative management mainly consists of medical blockade of gastric acid with proton pump inhibitors and total enteral feeding via percutaneous gastrostomy to unburden the esophageal lesion [5]. In addition, antibiotic treatment is applied in cases complicated by mediastinitis. Nevertheless, despite all these efforts, outcome of conservative management is almost invariably fatal due to recurrent hemorrhage or chronic mediastinitis [5]. Although our patient survived under conservative management for 14 months after the initial bleeding event, we will schedule the operative management of AEF if we observe recurrent bleeding episodes or signs of mediastinitis.

**CONCLUSIONS**

In conclusion, secondary AEF following endovascular stent-graft repair of the thoracic aorta is an uncommon but grave complication with only very limited therapeutic options. Although we observed 14 months survival in our case under

conservative management of secondary AEF, it seems that the treatment of secondary AEF following endovascular stent-graft repair of the thoracic aorta should do the operative management, considering the previous report [5] described about the risk of conservative management.

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