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CASE REPORT

# Unexpectedly large aortoesophageal fistula inconsistent with CT imaging due to the thrombus working as the tamponade

**Naoto Fukunaga (MD)[a,*], Takashi Matsueda (MD)[a], Masahiro Osumi (MD)[a], Atsushi Kurushima (MD)[a], Takashi Otani (MD)[a], Yoshiaki Fukumura (MD)[a], Shinobu Hosokawa (MD)[b], Yoshikazu Hiasa (MD, PhD, FJCC)[b]**

[a] *Department of Cardiovascular Surgery, Tokushima Red Cross Hospital, 103 Irinoguchi, Komatsushima-cho, Komatsushima-shi, Tokushima 773-8502, Japan*
[b] *Department of Cardiology, Tokushima Red Cross Hospital, Tokushima, Japan*

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**KEYWORDS**
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**Summary** A 69-year-old woman with a history of graft replacement of descending aortic aneurysm was referred to our hospital due to massive hematemesis with shock status. Additionally, the deterioration of respiratory status made us start the management under mechanical ventilation. The emergent gastrointestinal endoscopy by a general practitioner showed ulcer-like lesion of the upper esophagus and arterial bleeding. A contrast-enhanced computed tomography showed thoracic aortic aneurysm surrounded by low density mass and contrast medium protruding from the aneurysm. The findings suggested that thoracic aortic aneurysm perforated into esophagus and made an aortoesophageal fistula. Hemodynamic deterioration rapidly progressed and she passed away 4 days after her hospitalization.

Autopsy showed that a new thoracic aortic aneurysm arose from the proximal site of the graft. The aneurysm ruptured to esophagus with 6.0 cm × 5.0 cm sized fistula. The fistula was filled with a large thrombus. The large thrombus filling with the fistula worked as the tamponade and prevented her from the fatal exsanguinations and sudden death. The mechanism of the sentinel bleeding and the fatal exsanguinations known as Chiari's triad was revealed in our report.


## 1. Case report

A 69-year-old woman, who had a history of graft replacement of a descending aortic aneurysm 8 years previously was referred to our hospital

* Corresponding author. Tel.: +81 8853 2 2555; fax: +81 8853 6350.
*E-mail address:* naotowakimachi@hotmail.co.jp (N. Fukunaga).

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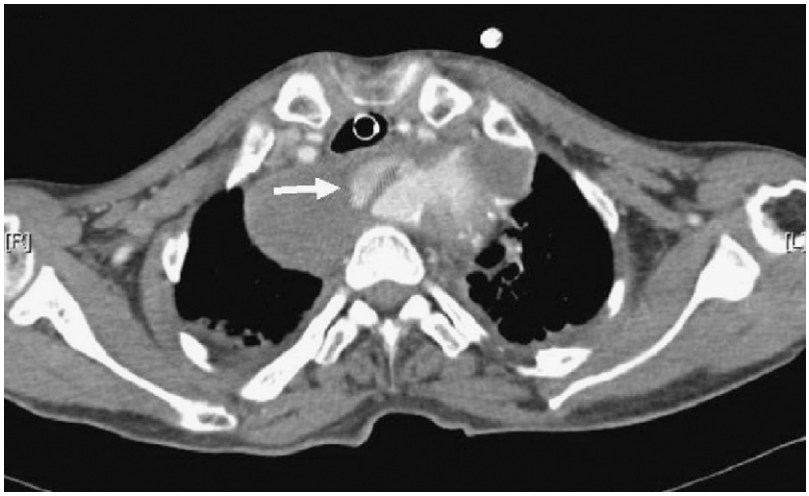

**Fig. 1** Enhanced axial computed tomography on admission. The arrow shows contrast medium protruding from the aneurysm. The esophagus could not be recognized and the bronchus deviated from original to anterior portion due to the compression of the aneurysm.

due to massive hematemesis. Her awareness was semicoma. She was in a cold sweat and cyanotic. Initial vital signs showed blood pressure of 66 mmHg/45 mmHg, pulse rate of 143 per minute, and a respiratory rate of 25 per minute. Because the respiratory status worsened gradually, management was initiated under mechanical ventilation after endotracheal intubation at an emergency department.

The laboratory findings showed hemoglobin level of 6.5 g/dl, white blood cell count of 12,410/ml with 72.4% neutrophils, platelet count of $6.4/\text{ml} \times 10^4/\text{ml}$, blood urea nitrogen of 21 mg/dl, creatinine of 1.32 mg/dl, and potassium of 4.2 mEq/l.

Emergent gastrointestinal (GI) endoscopy by a general practitioner showed ulcer-like lesion of the upper esophagus and arterial bleeding. A contrast-enhanced axial computed tomography (CT) on admission showed thoracic aortic aneurysm surrounded by low density mass and contrast medium protruding from the aneurysm (Fig. 1). The esophagus could not be recognized due to the compression of the aneurysm. Moreover, the bronchus deviated to anterior portion for the same reason. These findings suggested that thoracic aortic aneurysm perforated into esophagus and made an aortoesophageal fistula. During her hospitalization, intensive treatment continued under mechanical ventilation with blood transfusion.

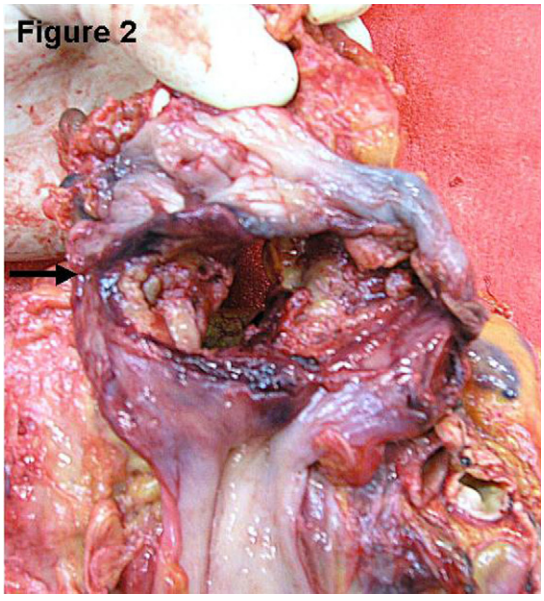


**Fig. 2** Photograph from the aspect of the esophagus incised longitudinally at autopsy. The arrow shows the 6.0 cm × 5.0 cm sized aortoesophageal fistula.

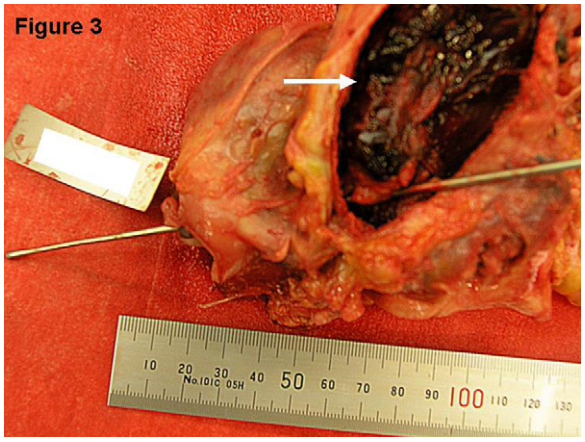


**Fig. 3** Photograph of the fistula at autopsy. The arrow shows a large thrombus recognized within the fistula.

Although surgical repair or stent graft was considered to improve the patient's general status, hemodynamic deterioration rapidly progressed and she passed away 4 days after her hospitalization.

Autopsy showed that a new thoracic aortic aneurysm arose from the proximal site of the previous graft. The aneurysm ruptured to the esophagus with 6.0 cm × 5.0 cm sized fistula (Fig. 2). The fistula was filled with a large thrombus (Fig. 3). The low density mass shown on CT turned out to be this thrombus.

## 2. Discussion

The first case of aortoesophageal fistula (AEF) was reported by Dubrueil in 1818 [1]. The patient he supported after the ingestion of a foreign body died due to hematemesis and autopsy revealed a fistula between the thoracic aorta and the esophagus. AEF is rare (0.01—0.08%) [2], but it is a life-threatening condition. It is classified into 2 groups, primary and secondary. Primary AEF is a very rare complication, with incidence ranging from 0.04 to 0.07% [3]. Thoracic aortic aneurysm is the most common cause (54%) of AEF. Foreign body ingestion such as needles and fish bones, advanced esophageal carcinoma, endoscopic procedure, trauma and radiotherapy are also causes of AEF [2]. An aneurysm can produce esophageal perforation by chronic stimulation related to erosion, inflammation, and necrosis. Secondary AEF, with an incidence of about 5% [2], is known as a complication due to previous aortic or esophageal surgery. At autopsy, we confirmed a new thoracic aortic aneurysm arising from the proximal site of the graft and the fistula between this new aneurysm and esophagus. We assumed that neither the previous surgery nor a pseudoaneurysm by the surgery affected the occurrence of this fistula, rather it was formed by a true aneurysm arising newly regardless of the previous surgery. This point was interesting enough to present this case.

Chiari's triad described in 1914 [4] suspicious symptoms of AEF, which included chest pain, sentinel bleeding, and fatal exsanguinations after a symptom-free period. The sentinel bleeding is caused by the rupture of the aortic aneurysm through a fistula into the nearby esophagus anatomically. It is minor or intermittent bleeding due to the thrombus formed within the fistula as a result of hypotension. In short, the thrombus works like a tamponade [5]. Additionally, the fistula becomes larger with repeated dislodgement of clots [6]. When the patient has high blood pressure, the thrombus will be pushed out from the fistula and subsequently the fatal exsanguination will occur. This interval between the sentinel bleeding and the fatal exsauguination ranges from hours to months [7]. Since there is only a little time left and the mortality rate is very high, prompt diagnosis and surgical intervention for aortic aneurysm are needed [2].

Regarding the outcome of surgical intervention, depending on the size of AEF and whether gross contamination is present, esophagectomy eliminating new infectious source of the mediastinum and prosthetic graft [6,8] and in situ replacement of the aortic aneurysm using prosthetic graft are the preferable procedures after debridement and removal of devitalized tissues, subsequently, abundant irrigation of mediastinum and protection of the prosthesis using variable pedicle flap such as the greater omentum [9]. Without gross contamination, primary closure of the esophageal fistula may be possible [2].

In recent years, percutaneous endovascular techniques for thoracic aortic aneurysm have increasingly developed. For high-risk patients, it is one alternative procedure instead of surgery. Within the limits of AEF, this technique may cause adverse events, because the esophageal fistula is left and the fistula contacting with the stent graft causes an inflammatory reaction, with subsequent periaortic infection and mediastitis. Therefore, in cases of AEF, endovascular techniques should be regarded as the procedure to stop the bleeding from the aorta and the bridge to second-staged procedure such as in situ replacement of the aortic aneurysm using prosthetic graft and esophagectomy removing the infectious source [2].

With regard to diagnosis, endoscopy is a useful modality to detect the origin of the GI bleeding and rule out the differential diagnoses such as esophageal varices and upper GI ulcer, leading to GI bleeding. CT has characteristic findings including contrast penetrating into the bowel, air within aortic wall, or focal bowel wall thickening [3]. Some reports said that CT did not demonstrate the fistula [2]. In our case, contrast medium protruding from the aneurysm was recognized on CT. Indeed esophagus was compressed by the aneurysm and not recognized on axial CT, but we predicted the presence of the fistula between aorta and esophagus and that the size of the fistula was small judging from the protruding contrast medium imaging on CT. However, the 6.0 cm × 5.0 cm sized AEF was recognized at autopsy, whose size was inconsistent with that we predicted from protruding contrast medium on CT. Moreover, regardless of the large fistula, this patient did not have fatal exsanguination to the last. As the mentioned mechanism above, the large thrombus filling with the fistula worked as the tamponade and prevented her from fatal exsan-

guination and sudden death [5]. In some cases, a large sub-epicardial hematoma and subsequent cardiac tamponade caused the hypotension and acute global left ventricular dysfunction [10]. In contrast with our case, the tamponade of that case deteriorated the patient's condition.

In conclusion, the mechanism from the sentinel bleeding to the fatal exsanguinations known as Chiari's triad was revealed in this report. From this point, we learned that we could not predict the size of the fistula judging from images, but could predict the presence of the fistula. Therefore, it is important to diagnose this condition from the images without underestimation and to perform surgical intervention immediately.

## References

[1] Dubrueil. Observations sur la perforation de l'oesophage et de l'aorte thoracique par une potion d'os avale: avec des reflexions. J Univ Sci Med 1818;9:357—63.
[2] Pirard L, Creemers E, Damme HV, Laurent S, Honor'e P, Limet R. In situ aortic allograft insertion to repair a primary aortoesophageal fistula due to thoracic aortic aneurysm. J Vasc Surg 2005;42:1213—7.
[3] Song Y, Liu Q, Shen H, Jia X, Zhang H, Qiao L. Diagnosis and management of primary aortoenteric fistula—experience learned from eighteen patients. Surgery 2008;143: 43—50.
[4] Chiari H. Ueber Fremdkorpeverletzung des Oesophagus mit Aortenperforation. Berlin Klin Wochenschr 1914;51:7—9.
[5] Guinn GA, Garcia-Rinaldi R, Whisennand HH. Surgical management of the bleeding aortopulmonary fistula. J Thorac Cardiovasc Surg 1976;71:554—6.
[6] Raghavendran K, Cherr GS, Ford PF, Burkhard PG, Bell-Thomson J. Successful management of concomitant aortoesophageal and aortotracheal fistulae secondary to a thoracic aortic aneurysm: case report and review of literature. J Vasc Surg 2005;42:1218—20.
[7] Saers SJ, Scheltinga MR. Primary aortoenteric fistula. Br J Surg 2005;92:143—52.
[8] da Silva ES, Tozzi FL, Otochi JP, da Tolosa EMC, Neves CRB, Fortes F. Aortoesophageal fistula caused by aneurysm of the thoracic aorta: successful surgical treatment, case report, and literature review. J Vasc Surg 1999;30:1150—7.
[9] Coselli JS, Crawford ES. Primary aortoesophageal fistula from aortic aneurysm: successful surgical treatment by use of omental pedicle graft. J Vasc Surg 1990;12: 269—77.
[10] Kashiwase K, Ueda Y, Ogasawara N, Oyabu J, Okada K, Murakawa T, Hirayama A, Kodama K. A large dissecting sub-epicardial hematoma and cardiac tamponade following elective percutaneous coronary intervention. J Cardiol 2008;52:163—6.