Turk J Gastroenterol *2014; 25 (Suppl.-1): 321-3*

Aortoesophageal fistula (AEF): Fatal upper gastrointesinal haemorrhage

To the Editor,

We present the case of a 42-year-old male diagnosed with Aortoesophageal fistula at autopsy, who was misdiagnosed as having a gastroduodenal ulcer associated with upper gastrointesinal haemorrhage. Unfortunately, medical and surgical treatment was unsuccessful and the patient died. Rapid diagnosis and immediate intervention are essential to reduce mortality among individuals with this condition.

Aortoesophageal fistula (AEF) is a rare but catastrophic disease where a fistula forms through the aorta and esophagus due to various causes, which are characterized by a triad of mid-thoracic pain or dysphagia followed by a "herald" hemorrhage and fatal hematemesis. It is extremely difficult to diagnosis AEF in a timely manner due to the rapid progression after upper gastrointesinal haemorrhage. We report a case of AEF which was diagnosed at autopsy and discuss the etiology, diagnosis, treatment of AEF to highlight the challenges associated with identifying this lethal condition.

A 42-year-old male was given a preliminary diagnosis of upper gastrointesinal haemorrhage because of vomiting bright red blood combined with a 2-day history of melaena. Upper gastrointestinal endoscopy revealed a lot of fresh blood and clots deposited in the fundus gastricus and corpus gastricum, which appeared to initially come from the esophagus, however, the source of the bleeding could not be identified. Approximately one hour following endoscopy, the patient experienced two successive episodes of hematemesis, with a total volume of approximately 500 ml. Emergency exploratory laparotomy found that the stomach was expanded and filled with large amounts of fresh blood and clots, nevertheless, the area of active bleeding was not detected. An empirical partial gastrectomy and gastrojejunostomy was undertaken in order to control upper gastrointesinal haemorrhage. However, within one hour after the procedure there was another episode of hematemesis. As anastomotic stoma bleeding was suspected, an emergency exploratory laparotomy was undertaken once more, which found that the stomach was occupied with a lot of bright red blood while the anastomotic stoma had not been lacerated or bleeding. It was suspected that the origin of bleeding may have come from the inferior segment of the esophagus, therefore, given the deteriorating state of the patient, it was decided to inflate the esophagus with a Sengstaken-Blakemore tube to control bleeding. Unfortunately, the patient died due to cardiopulmonary arrest half an hour after the operation. A week later, autopsy revealed a thoracic aortic dissecting aneurysm which was located at the 6-8th thoracic vertebrae, part of which was calcified. The aortic aneurysm wall had a round rictus adhered by an organized thrombus, which communicated with the swollen esophagus. Therefore, the postmortem diagnosis of the patient was AEF caused by the thoracic aortic dissecting aneurysm (Figure 1).

AEF is an incredibly dangerous occult condition that is usually fatal if not diagnosed early. However, it is also reasonable that AEF is usually defined as the final diagnosis when the salient features of this case in which the source of bleeding was not coming from the stomach but esophagus was considered, because 80% of AEF cases have a sentinel hemorrhage prior to fatal exsanguination (1).

Cautious esophagoscopy is the most sensitive and specific diagnostic technique for diagnosing AEF (2), AEF should be suspected if there are massive hematemesis but endoscopy shows little blood in the stomach and no obvious source (1). Computed tomographic scanning although does not demonstrate the fistula, but it can delineate the location of an aneurysm and its surrounding structures, especially of the esophagus, which is extremely valuable for surgical planning.

Address for Correspondence: Yi-hu Fang, Department of Pathology, Shangrao Branch of Jiangxi Medical College, Shangrao, China
E-mail: fyh3101@sohu.com
Received: November 17, .2012 **Accepted:** February 28, 2013
 • Available online at www.turkjgastroenterol.org • DOI: 10.5152/tjg.2014.4544

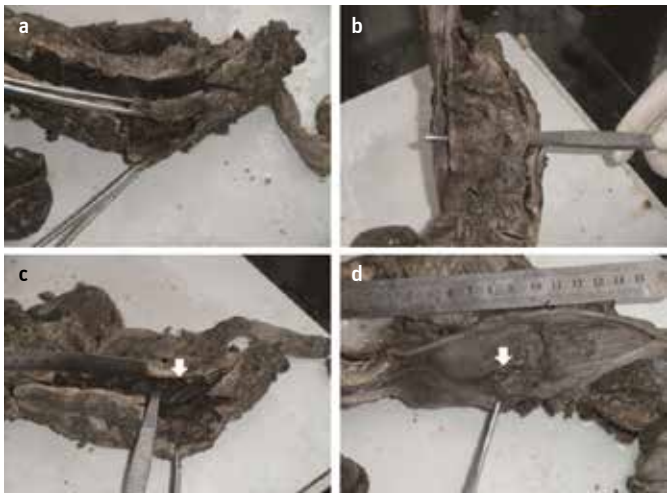

Figure 1. (a,b,c,d). Image of an AEF. **(a)** thoracic aortic dissecting aneurysm. **(b)** AEF. **(c)** the aortic side of AEF (*white arrow*). **(d)** the esophageal side of AEF (*white arrow*).

Surgery is the only definitive treatment option for AEF and should be initiated as soon as possible. There are a number of challenges faced by surgeons including control of hemorrhage, arterial reconstruction, control of sepsis, and re-establishment of the alimentary tract (3). Replacement of the affected aorta with a cryopreserved arterial allograft is usually the procedure of choice (2). The prognosis of AEF surgical treatment depends on the size of AEF and whether debridement is complete, currently, there is no consensus on whether primary repair or esophageal resection is the better treatment option for esophageal tear associated with aortoesophageal fistula. Regardless of the fact that some reports recommend primary repair of the esophagus, leakage from the repair site is a frequently reported complication (4). Subtotal esophageal resection, followed by gastric or colonic interposition, could significantly minimize the risks of graft infection and seems to be the most promising solution (5).

Due to the rapid development of minimally invasive techniques, endovascular techniques have recently gained acceptance for the treatment of AEF. Verhoeven suggested that endovascular aortic stent grafts should be considered as the first-choice lifesaving procedure.6 A combination of surgical and endoluminal stent or esophageal and aortic stents to treat AEF can achieve a beneficial effect (7,8). However, endovascular techniques have some limitations such as the AEF remaining as it is, insufficient debridement or drain of the mediastinum, which can increase the rise of septic complications, furthermore, if the endograft collapses during the treatment of an aortoesophageal fistula, the consequences would be disastrous (9). Therefore, some studies have suggested that the endovascular repair may be only suitable for patients with a high risk of open surgery or for the temporary control of bleeding. However, recent reports suggest that aortic endovascular stent graft and retrievable esophageal stent were placed followed by thoracoscopic mediastinal debridement and drained successfully to treat a patient with AEF (10), which means that endovascular repair treatment of AEF can break the bottleneck of its limitations.

In conclusion, AEF is an uncommon but life-threatening disease which should be taken into consideration when unexplained massive upper gastrointestinal haemorrhage has occurred, only rapid diagnosis and immediate intervention will reduce morbidity and mortality. Computed-tomography is a useful diagnostic test to find AEF. Endovascular repair may be the best way to treat AEF, however, these methods will require further evaluation with larger studies that include long-term follow-up of patients. In this case, the exact location of bleeding was not clear, however, if the patient had a CT examination performed during the stabilization period after endoscopy, this may have assisted in locating the source of the bleeding and provided a clear diagnosis of esophageal aortic fistula, interventional endovascular techniques could have been applied, which may have saved the patient.

Conflict of Interest: No conflict of interest was declared by the authors.

Yi-hu Fang[1], Xue-liang Zhou[2], Ji-chun Liu[2]

[1]Department of Pathology, Shangrao Branch of Jiangxi Medical College, Shangrao, China

[2]Department of Cardiothoracic Surgery, The First Affiliated Hospital, Nanchang University, Nanchang, China

REFERENCES

1. Amin S, Luketich J, Wald A. Aortoesophageal fistula: case report and review of the literature. Digestive diseases and sciences 1998; 43: 1665-71.
2. Pirard L, Creemers E, Van Damme H, Laurent S, Honore P, Limet R. In situ aortic allograft insertion to repair a primary aortoesophageal fistula due to thoracic aortic aneurysm. Journal of vascular surgery : official publication, the Society for Vascular Surgery [and] International Society for Cardiovascular Surgery, North American Chapter 2005; 42: 1213-7.
3. Baril DT, Carroccio A, Ellozy SH, Palchik E, Sachdev U, Jacobs TS, et al. Evolving strategies for the treatment of aortoenteric fistulas. Journal of vascular surgery : official publication, the Society for Vascular Surgery [and] International Society for Cardiovascular Surgery, North American Chapter 2006; 44: 250-7.
4. Tokuda Y, Matsumoto M, Sugita T, Nishizawa J, Matsuyama K, Yoshida K, et al. Successful repair of an aortoesophageal fistula caused by a thoracic aortic aneurysm: report of a case. Surgery today 2004; 34: 357-9.
5. Yamanaka K, Nonaka M, Iwakura A, Asao Y. Repair of aortoesophageal fistula after total aortic arch grafting. Interactive cardiovascular and thoracic surgery 2011; 12: 655-6.
6. Verhoeven EL, Vourliotakis G. Thoracic endovascular aortic repair for aortobronchial or aortoesophageal fistulas: permanent or temporary salvage or not an option at all? Journal of endovascular therapy : an official journal of the International Society of Endovascular Specialists 2009; 16: 441-2.
7. Marone EM, Coppi G, Kahlberg A, Tshomba Y, Chiesa R. Combined endovascular and surgical treatment of primary aortoesopha-

geal fistula. Texas Heart Institute journal / from the Texas Heart Institute of St Luke's Episcopal Hospital, Texas Children's Hospital 2010; 37: 722-4.

8. Ghosh SK, Rahman FZ, Bown S, Harris P, Fong K, Langmead L. Survival following Treatment of Aortoesophageal Fistula with Dual Esophageal and Aortic Intervention. Case reports in gastroenterology 2011; 5: 40-4.
9. Zaporteza K, Anaya-Ayala JE, Davies MG, Peden EK, Lumsden AB. Recurrent collapse of a Gore TAG endograft in treating an aortoesophageal fistula. Vascular 2011; 19: 154-8.
10. Chen X, Li J, Chen J, Zhou Y, Zhang Y, Ding H, et al. A combined minimally invasive approach for the treatment of aortoesophageal fistula caused by the ingestion of a chicken bone: case report and literature review. Clinics (Sao Paulo) 2012; 67: 195-7.