The pulsatile bulge on the oesophagus: rare cause for hematemesis

S.Vinojan¹, S. Mathievaanan², S. Giridaran³, D. Karunarasan¹, S. Ratnajothy¹
¹University of Jaffna, Sri Lanka
²Teaching Hospital Jaffna, Sri Lanka

Keywords: Aortoesophageal fistula; mycotic aneurysm; pulsatile bulge

Introduction
Aortoenteric fistula is a rare but fatal condition. Here we present a case of primary aortoenteric fistula due to a thoracic aneurysm.

Case presentation
A 52 years old gentleman was admitted with a history of chest pain for 3 weeks duration associated with dysphagia. Although he denied a history of fever, he had a history of loss of appetite and loss of weight. The initial examination was unremarkable.

His white cell count was 22,000/mm³ with 90% of neutrophils, haemoglobin was 10g/dL and platelet was 364 X 10⁹/L. His C-Reactive Protein (CRP) was 221 mg/L on admission. Other blood investigations were unremarkable. The initial blood culture was negative and the echocardiogram did not reveal any vegetation. As he had a history of dysphagia, he underwent upper gastrointestinal endoscopy (UGIE) which revealed a narrowing of the oesophagus at the level of 28 cm from incisor teeth but there were no mucosal abnormalities. Subsequently, he underwent Computed Tomography (CT) chest and abdomen. CT revealed a saccular descending thoracic aneurysm extending up to the level of the celiac artery with evidence of concealed rupture (Figure 1) and gas pockets around the aneurysm (Figure 2). While he was awaiting an urgent repair, he developed massive hematemesis and died.

Discussion
Aortoenteric fistula (AEF) is a rare but highly fatal disease. Dubreuil described this condition in 1818 in a patient who ingested beef rib [1]. AEF can be primary or secondary. Primary AEF occurs in the native aorta due to aneurysm, foreign body, malignancy and radiation. Secondary AEF occurs in an aorta which was previously operated [2].

Correspondence: Satchithanantham Vinojan
E-mail: vinojan@univ.jfn.ac.lk
https://orcid.org/0000-0002-7172-997X
Received: 20-12-2021   Accepted: 06-07-2022
DOI: http://doi.org/10.4038/sljs.v40i2.8928

The Sri Lanka Journal of Surgery 2022; 40(2): 39-40
Oesophagus is affected in 28-30% of cases of aortoenteric fistula [2]. Chiari explained the triad of symptoms; mid-thoracic chest pain or dysphagia, sentinel haemorrhage followed by massive haemorrhage [3]. Our patient had this typical triad although only 45% of the patients with AEF presented with Chiari’s triad [4]. Initial mid thoracic pain may be due to rapid stretching of the aortic wall, erosion and dissection of the aortic wall and oesophageal perforation. Air pockets in the CT chest can be due to mediastinitis due to oesophageal perforation which is also another cause of chest pain. Dysphagia was due to mechanical compression. Initial herald bleeding settled because of hypotension and clot formation.

UGIE and computed tomography angiogram (CTA) can be used to diagnose the fistula. There are reported cases where initial UGIE might fail to diagnose AEF as in our case. Pulsatile bulging with or without adherent clots is rarely seen in UGIE [2].

In patients with hematemesis and aortoesophageal fistula, thoracic stenting would be a bridging therapy or primary treatment. In long run, there is a chance of stent graft infection due to a mycotic aneurysm, so some authors recommend lifelong antibiotic treatment. If the stenting is done as bridging therapy aorta should be replaced with either a prosthetic graft or an autologous graft. A prosthetic graft carries the same risk of infection as a stent graft. There are reported cases of replacement of aorta with bovine pericardium [5]. In this patient, as he developed massive hematemesis immediately after the diagnosis we couldn’t proceed with stenting or definitive surgical repair.

Aortoenteric fistula should be suspected in patients with mid-thoracic pain and herald bleeding which is bright red. Unnecessary delay in initial diagnosis might negatively affect the outcome.

**Conclusion**

Even though upper gastrointestinal bleeding due to aortoenteric fistula is rare, it should be suspected in a patient with chest pain and dysphagia. Initial herald bleeding is the only warning sign and thoracic covered stenting would be the ideal treatment at least as a bridge.

All authors disclose no conflict of interest. The study was conducted in accordance with the ethical standards of the relevant institutional or national ethics committee and the Helsinki Declaration of 1975, as revised in 2000.

**References**

1. Kokatnur L, Rudrappa M. Primary aorto-esophageal fistula: great masquerader of esophageal variceal bleeding. Indian journal of critical care medicine: peer-reviewed, official publication of Indian Society of Critical Care Medicine. 2015 Feb;19(2):119. doi: 10.4103/0972-5229.151022

2. Monteiro AS, Martins R, da Cunha CM, Moleiro J, Patrício H. Primary Aortoesophageal Fistula: Is a High Level of Suspicion Enough? European Journal of Case Reports in Internal Medicine. 2020;7(8). doi: 10.12890/2020_001666

3. Chiari H. Ueber Fremdkorperverletzung des oesophagus mit aortenperforation. Ber Klin Wochenschr. 1914;51:7-9

4. Heckstall RL, Hollander JE. Aortoesophageal fistula: recognition and diagnosis in the emergency department. Annals of emergency medicine. 1998 Oct 1;32(4):502-5. doi:10.1016/S0196-0644(98)70182-9

5. Al - Thani H, Wahlen B M, El - Menyar A, Hussein A, Sadek A, Fares A, Sainh MM, Almulla A. Presentation, management and outcome of aorto-esophageal fistula in young patients: two case-reports and literature review. Journal of Surgical Case Reports. 2021 Jun;2021(6): rjab213. doi: 10.1093/jscr/rjab213

**Learning Points:**

- Primary aortoenteric fistula is a rare cause of hematemesis
- Typical triad of chest pain, sentinel haemorrhage followed by massive haemorrhage only occur in some patients.