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Aorto-Oesophageal Fistula; A Rare Cause of Acute Upper Gastrointestinal Bleeding. A Case Report

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Authors' contributions

This work was carried out in collaboration between both authors. Both authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Aorto-oesophageal fistula is a considerably rare but potentially fatal cause for upper gastro-intestinal bleeding. Mortality is high due to delayed diagnosis, delayed treatment due to limited facilities at the first contact health care centres and the possibility of re-bleeding even after successful initial treatment. The classic clinical picture has 3 symptoms termed Chiari's triad which include mid thoracic pain, sentinel arterial haemorrhage and exsanguination. Diagnosis could be made by the clinical picture alone. Most investigations aimed at diagnosis have low specificity and sensitivity. The most frequently used test is upper GI endoscopy for diagnostic as well as treatment purposes. Definitive treatment is left thoracotomy and fistula repair. This is a case report of a 61-year-old male who died due to aorto-oesophageal fistula even after upper GI endoscopy had been performed.

Keywords: Thoracotomy; upper GI endoscopy; chiari's triad; aortic aneurysm; ischaemic heart disease.

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1. INTRODUCTION

Upper gastrointestinal (GI) bleeding is a common surgical condition all over the world. Ruptured esophageal varices, esophageal carcinomas, eroding peptic ulcers, Mallory-Weiss tears, surgical interventions are some common causes for upper GI bleeding. Acute upper gastrointestinal bleeding may be a potentially life threatening condition requiring prompt and proper medical and surgical intervention. Apart from the above mentioned common conditions of upper GI bleeding, there could be few remote and rare causes for this clinical presentation. Aorto-oesophageal fistula is one such rare condition which was first recognized in 1818 [1]. Aorto-oesophageal fistulae may be primary or secondary. Primary aorto-oesophageal fistula is a rare entity with an incidence of 0.04-0.07%. Primary aorto-oesophageal fistulae are caused by atheromatous aortic aneurysms or less frequently by penetrating/eroding aortic ulcers. British surgeon and anatomist, Sir Astley Cooper described a typical case of aorto-oesophageal fistula in 1829. The majority of fistulae are of secondary type and could occur in a setting of prior aortic reconstructive procedure. Pathology of aorta is represented in 80% of all cases of primary aorto-oesophageal fistulae. The etiology of primary aorto-oesophageal fistula is uncertain but proposed theories include direct wear and inflammatory destruction triggered by infection, foreign bodies and erosion. The clinical presentation of aorto-oesophageal fistula is typically characterized by the so called Chiari's triad consisting of thoracic pain followed by herald bleeding, a short symptom-free interval and finally a fatal exsanguination.

2. CASE REPORT

A 61-year-old male had complained of loss of appetite and dysphagia for over two months. He had undergone an upper GI endoscopy in a state sector tertiary care hospital one early morning and had been discharged the same afternoon. He developed chest pain and haematemesis the same evening at home. He collapsed on the way to hospital and was pronounced to be dead on admission. Post mortem examination revealed pale conjunctivae, pale organs, flame shaped sub-endocardial haemorrhages on the left ventricular wall and un-clotted blood in the stomach (approximately 650ml in volume) but no evidence of melena. Esophageal mucosa was free of acute injury and no evidence of ruptured esophageal varices was found. An ulcerative lesion of the upper esophagus measuring 3.8 cm x 3.5 cm with a fistula tract connecting the esophageal lesion with the superior part of the descending aorta was evident (Figs. 1 and 2). The thoracic and to a larger extent the abdominal aortic intima was studded with fissured, calcified and ulcerated atheromatous plaques. They were present around the fistulous connection on the aortic side. Both renal artery openings as well as the renal arteries themselves were extremely narrowed with complicated atheroma. Both kidneys appeared granular and contracted. There were more than 90% atheromatous narrowing of all three major coronary artery branches. The cause of death was given as haemorrhagic shock following aortic aneurysmal rupture into oesophagus.

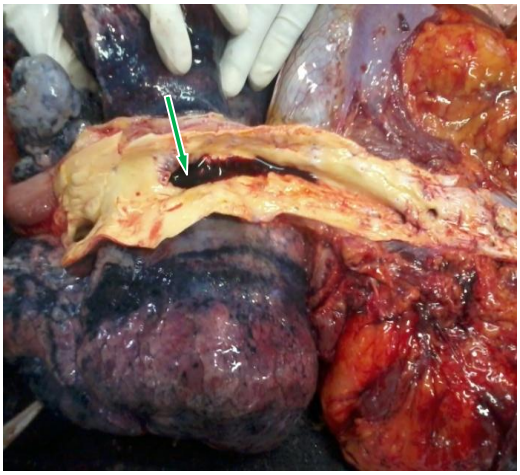

Fig. 1. Aortoeosphageal fistula

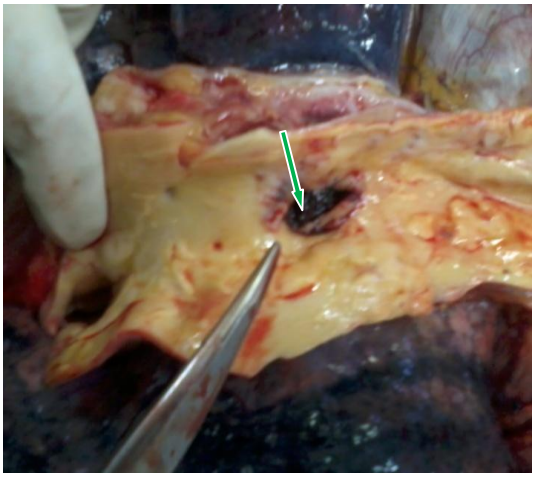

Fig. 2. Close-up view from aortic side

3. DISCUSSION

Primary aorto-oesophageal fistula is an uncommon, but life threatening cause of gastrointestinal bleeding, which often causes death by massive exsanguination [2]. Transient self-limited bleed called 'herald bleed' may precede fatal exsanguination and is often a significant feature of disease process. This may allow an opportunity for diagnosis and appropriate life-saving management. Diagnostic approach may include CT scan, upper GI endoscopy, RBC scan and angiography but no single modality has good sensitivity and specificity to arrive at a definitive diagnosis [3,4]. Chiari's triad is characteristic in a patient with aorto-oesophageal fistula, but it is only present in one third of the cases with this condition [5]. Prior diagnosis could be made by clinical assessment in a patient with classical symptoms and signs. Yet, according to a case series studied by Vaideeswar P, most of them were diagnosed only at autopsy [6]. A pulsating submucosal mass is typical and blood clots may obscure the exact site of fistula-formation, making the diagnosis more challenging. Surgical intervention is the definitive treatment for aorto-oesophageal fistula though the mortality rate is objectionably high [7]. Until the definitive treatment is done, bleeding may temporarily be arrested with Sangstaken-Blakemore tube. It should be used only if the patient is at risk of exsanguination. The diagnosis is mostly based on high clinical suspicion supplemented by judicious use of endoscopy. Even if the bleeding is stopped with endoscopy there is a considerable chance of re-bleeding with very high mortality. Though the aorto-oesophageal fistula is a rare cause for upper GI bleeding, it is a considerable cause in an elderly hypertensive patient. Post-operative aortic disease is the most common secondary cause [8,9]. There are other causes like oesophageal carcinoma eroding into an artery and Boerhaave syndrome both of which are relatively rare [10]. Another rare case in a relatively younger patient is foreign body ingestion like button batteries and fish bones which may erode the upper oesophagus and the adjacent aortic wall to form a fistulous track [11,12,13.

There are numerous investigations related to aorto-oesophageal fistula. Yet, there is no single definitive diagnostic method available. Upper GI endoscopy is the commonest method used for diagnosis of esophageal pathology. It is frequently accompanied with taking biopsy for histopathology. Biopsy might trigger the bleeding in a patient with aorta-oesophageal fistula as necrotic areas are more fragile. Such areas are more susceptible for rupture too. Vital reactions are not much prominent in those areas following trauma. Histological samples don't show tissue reaction related to the injury frequently. Taking biopsy may damage the area of the esophagus which is already eroded by aortic atheroma/aneurysm. Non-invasive investigative methods are recommended prior to the invasive procedures in a patient with features highly suggestive of aorto-oesophageal fistula. In the deceased under discussion here, the upper GI endoscopy report did not mention anything about the abnormal finding in the upper oesophagus which was approximately 3.8 x 3.5 cm in dimensions. The oesophageal margins did not show evidence of obtaining punch biopsy samples. The subsequent bleeding in this case might have been precipitated merely due to the mechanical manipulation of the oesophagus during the upper GI endoscopy or it might not have had any relationship with the procedure at all.

4. CONCLUSION

Aorto-oesophageal fistula is a rare cause of upper gastrointestinal bleeding. Most cases are due to atheromatous degeneration. The possibility of aorto-oesophageal fistula should be born in mind in patients presenting with thoracic pain and upper GI bleeding. Definitive treatment is surgical. When a fistula is already present, mechanical manipulation of the oesophagus during upper GI endoscopy may precipitate bleeding.

CONSENT AND ETHICAL APPROVAL

As per international standard or university standard guideline patients consent and ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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