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E-mail: jzus@zju.edu.cn

Case Report:

Aortoduodenal fistula following aortic reconstruction of a pseudoaneurysm caused by stab wound 12 years ago

Jian-cang ZHOU[†1], Qiu-ping XU[1], Lai-gen SHEN[2], Kong-han PAN[1], Yi-ping MOU[2]

([1]*Department of Critical Care Medicine, Sir Run Run Shaw Hospital, School of Medicine, Zhejiang University, Hangzhou 310016, China*)

([2]*Department of General Surgery, Sir Run Run Shaw Hospital, School of Medicine, Zhejiang University, Hangzhou 310016, China*)

[†]E-mail: jiancangzhou@hotmail.com

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Abstract: Gastrointestinal bleeding due to aortoenteric fistula is extremely rare. Aortoenteric fistula is difficult to be diagnosed timely and entails a significant morbidity and mortality. Herein, we present an uncommon case of gastrointestinal bleeding caused by aortoduodenal fistula, which was a complication of a successful aortic reconstruction 4 months ago for an aortic pseudoaneurysm resulted from a stab wound 12 years ago. An urgent laparotomy confirmed an aortoduodenal fistula and repaired the defects in aorta and duodenum, but a prolonged shock led to the patient's death. In summary, early diagnosis and surgical intervention for aortoenteric fistula are vital for survival.

Key words: Aortoduodenal fistula, Aortic pseudoaneurysm, Aortic reconstruction, Stab wound
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INTRODUCTION

Aortoenteric fistula (AEF) is an extremely rare abnormal communications between the aorta and the bowel, most frequently resulting from prosthetic graft erosion or a complication of aortic aneurysm compressing the adjacent bowel. Rupture of the aorta through AEF into a closely adherent portion of the gastrointestinal (GI) tract may lead to GI bleeding, which may range from a minor and intermittent hemorrhage at its initial stages, to an life-threatening exsanguinations (Song *et al*., 2008; Saers and Scheltinga, 2005; Goshtasby *et al*., 2005). Despite advances in surgery and medical technology, these entities are still associated with significant morbidity and mortality for the patient; the mortality is as high as almost 100% if left untreated (Lemos *et al*., 2003). The very low incidence of AEF in general population makes this condition a very uncommon entity in the differential diagnosis of GI bleeding. Therefore, physicians and gastroenterologists in the emergency departments usually have insufficient awareness of this rare disorder (Song *et al*., 2008). Here, we present a unique case of a post-stab pseudoaneurysm existing for 12 years together with a retained dagger in the abdomen, which was successfully managed with repair of primary aortic defects. The patient died unfortunately 4 months later for catastrophic GI bleeding as a result of a secondary AEF. We will also review the literature on AEF with a comprehensive overview on major clinical characteristics and current management options, in order to improve the diagnostic accuracy and management efficiency for patients with AEF.

CASE REPORT

First admission at our hospital

A 33-year-old woman was admitted to our hospital on Mar. 1, 2007, due to abdominal mass for 4 months. She had been excellently healthy until 4 months earlier, when a pulsatile mass was found in her epigastric region. The mass was not accompanied with abdominal cramps, fever, hematochezia, or loss

of weight. A further ultrasonography revealed a pseudoaneurysm of abdominal aorta, with the size about 10.2 cm×7.0 cm×7.1 cm. She had a stab wound 12 years ago, but the detail of the wound and treatment was unavailable. She had given birth to two children by cesarean section and both of them were healthy.

On admission, her abdomen was flat and soft, and a 3-cm scar and a round pulsatile mass about 7 cm were found at right epigastric region just below the costal arch. Laboratory findings were unremarkable.

A contrast-enhanced computed tomography (CT) showed an aortic pseudoaneurysm around 10 cm×9 cm, which contained thrombosis and a metallic foreign body (Fig.1). A further angiography showed that the defect of aorta at the Th10 level was approximately 3 cm in diameter.

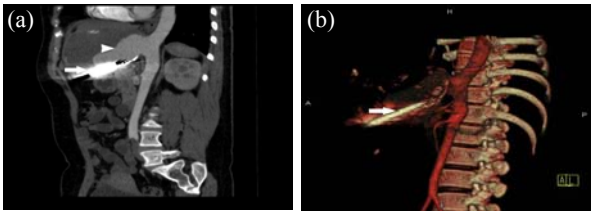

Fig.1 Contrast-enhanced computed tomography (CT) (a) and three dimension reconstruction (b) show pseudoaneurysm (arrowhead) contains a dagger (arrow), which is located between the stomach and liver

On Day 14, she underwent an aortic reconstruction surgery. A soft pulsatile mass measuring 10 cm×7 cm×7 cm was found between the gastric fundus and left hepatic lobe. Two balloons were placed at the two ends of the pseudoaneurysm and were inflated to occlude the blood flow in the aneurysm. There was no evidence of infection or abscess formation. A rotten dagger around 8 cm×3 cm and some clots were removed when the aneurysm was opened. The defect of aorta, which was about 3 cm×1.5 cm, was repaired in situ with prosthetic graft. The recovery was unevenly and she was discharged on Day 20.

Second admission at our hospital

The patient was readmitted on July 18, 2007, with intermittent hematemesis, hematochezia, and fever for 2 d. Her temperature varied between 38.2~39.2 °C and her hemoglobin level declined to as low as 49 g/L. On admission she was pale and hypovolemic shock. Urgent bedside upper GI endoscopy showed massive fresh blood in the duodenum and an ulcer around 0.4 cm×0.3 cm was found on the anteriolateral wall of superior part of the duodenum. Bleeding stopped when the artery in the ulcer was clamped and 1:10000 epinephrine was injected around the ulcer. The patient remained relatively stable for 2 d. On Day 3, the hematochezia appeared again. A second GI endoscopy found massive fresh blood in the stomach and duodenum but the source of the blood was not clear. Contrast enhanced CT revealed peri-aortic ectopic gas around the graft (Fig.2). The hemodynamic was unstable, requiring an epinephrine infusion to maintain mean arterial blood pressure. So, she underwent an urgent laparotomy surgery.

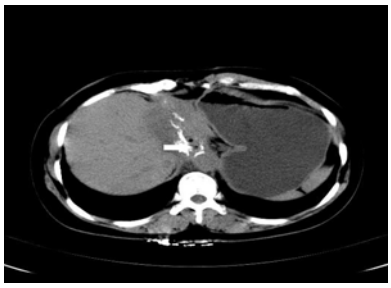

Fig.2 Computed tomography (CT) shows the presence of peri-aortic ectopic gas (arrow) around the graft

Severe adhesions were encountered in the peritoneal cavity. Bile staining and erosion of the graft were observed. Following a careful dissection of severely adhered tissues, a connection around 2.0 cm was identified between duodenal wall and abdominal aorta. The fistula was closed with new graft, and the defect in the duodenum was repaired. The patient's postoperative course was complicated with a prolonged shock and multiple organ dysfunctions, and she died 3 d later.

DISCUSSION

Traumatic injury of the abdominal aorta is rare but potentially lethal, as it can result in major hemorrhage, requiring an urgent open surgery. In current case, we present a 33-year-old woman with aortic pseudoaneurysm resulting from stab wound 12 years ago, who had successively given birth to two children

by cesarean section and survived even with an 8-cm dagger retained in her abdomen for 12 years.

An AEF is a potentially catastrophic clinical entity that can be categorized as primary or secondary according to the cause. Primary fistulas are sometimes seen in patients with an abdominal aortic aneurysm and atherosclerosis, whereas secondary fistulas are more common and are manifested as complications of previous aortic surgery (Graber *et al*., 2007). Time regarding the duration between the aortic surgery and AEF ranges from several days to more than 10 years (Busuttil and Goldstone, 2001; Baril *et al*., 2006).

AEF is difficult to diagnose, and a large proportion of patients with AEF die before a correct diagnosis is made due to its variable clinical manifestations, long interval after primary aortic surgery, and insufficient awareness of this rare entity in physicians and gastroenterologists (Saers and Scheltinga, 2005; Goshtasby *et al*., 2005). The expected triad of abdominal pain, pulsatile abdominal mass, and GI bleeding is encountered only in less than 25% cases (Saers and Scheltinga, 2005), which may lead to a delay in diagnosis. It is common for AEF to present initially with a minor, so-called "herald" bleeding, which is later followed by a catastrophic, life-threatening bleeding. The duration of herald bleeding and final exsanguination ranges from hours to months, with 40% more than one week (Voorhoeve *et al*., 1996). This duration provides valuable opportunity for surgical intervention.

Making the diagnosis of AEF requires a high index of suspicion, especially in patients with history of aortic reconstruction surgery. CT is the most useful technology in clinic practice nowadays. Signs such as perigraft ectopic gas and intravasation of contrast into the bowel lumen or perigraft space favor the diagnosis of AEF (Perks *et al*., 2004). A study has compared CT scan findings with operative results and concluded that this examination had a sensitivity of 94% and a specificity of 85% in detecting AEF (Hughes *et al*., 2007). Multidetector CT, providing excellent image quality and enabling volume-rendered image reconstruction, is particularly useful for the diagnosis of AEF when the patient's bleeding rate is slow and when detection of the proximal part of the fistula tract might be enough for diagnosis (Ödemiş *et al*., 2008). Upper GI endoscopy is the preferred procedure for the evaluation of upper GI bleeding, whatever the cause. GI endoscopic evaluation often identifies the site of bleeding and allows directed urgent lifesaving therapy. However, the sensitivity of upper GI endoscopy in diagnosis of AEF has been reported to be only 50% (Wood *et al*., 2005; Hughes *et al*., 2007). What's worse, findings of gastritis or peptic ulcer other than AEF itself by GI endoscopy may be misleading and lead to a delayed diagnosis of AEF (Saers and Scheltinga, 2005; Mavioglu *et al*., 2005). In the present case, findings of ulcers of the double upper GI endoscopy before laparotomy attracted doctors' attention too much, which led to the delayed diagnosis and treatment of the patient. Angiographic filming technology is useful to define arterial anatomy and to allow the planning of aortic reconstruction, and intravasation of contrast into the bowel lumen is the standard for the diagnosis of AEF. Whereas it is dangerous for AEF patients with recurrent or active bleeding to take angiography because high-pressure injection of contrast increases the possibility of a catastrophic bleeding (Wang *et al*., 2006).

Patients with AEF doomed to death before 1970's, and surgery is the only viable chance of a cure today (Saers and Scheltinga, 2005; Song *et al*., 2008). A review of 118 cases of AEF by Sweeney and Gadacs (1984) showed that the total and operation-related mortalities were 86% and 36%, respectively, and most of them died before diagnosis or surgical intervention. Early recognition of herald bleeding and urgent surgical intervention is essential to decrease the mortality rate of AEF. Some authors suggested to place an occlusive balloon at, or above, the level of the AEF, in order to gain time to consider different therapeutic options and even to transport the patient within the institution when needed (Leonhardt *et al*., 2008). Treatment typically entails removal of the graft and any infected tissue, revascularization, and closure of the bowel fistula. Various operative strategies have been reported for this complication, including in situ aortic graft replacement with a variety of new aortic grafts for those without gross infection, while axillobifemoral bypass is reserved only for patients with extensive local sepsis (Goshtasby *et al*., 2005). The advent of percutaneous endovascular techniques revolutionized the management of AEF, especially for patients unsuitable for open surgery. Danneels *et al*.(2006) reviewed 15 cases between

1999 and 2005 and concluded that endovascular sealing of AEF provides time to treat shock, local and systemic infection, and comorbidity, which creates a better situation to perform future open repair, with possibly better outcome.

In summary, early diagnosis and surgical intervention for AEF are crucial for patient survival. AEF must be kept in mind as feasible etiology of massive GI bleeding in patients with a known history of aortic reconstruction, no matter how later after previous operation.

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