A very Rare Complication of an Abdominal Aortic Aneurysm. Primary Aortoduodenal Fistula without Gastrointestinal Bleeding.

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Abstract

An abdominal aortic aneurism (AAA) is an enlargement of the lower part of the aorta that extends through the abdominal area. The diameter of the aneurismatic vessel is represented by 3 cm or more in either anterior – posterior, or transverse planes. Aortoenteric fistula (AEF) is defined as a communication between the aorta and gastrointestinal (GI) tract. We report the case of an 84-year-old man presenting with a two-day history of epigastric and back pain. The patient was diagnosed with AAA complicated with primary aortoduodenal fistula. This paper aims to describe the management of AAA and its complications as well as the review the actual literature.

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Full Text

Introduction
An abdominal aortic aneurism (AAA) is an enlargement of the lower part of the aorta that extends through the abdominal area. The diameter of the aneurismatic vessel is represented by 3 cm or more in either anterior-posterior, or transverse planes. The development of Abdominal aortic aneurysm (AAA) is a complex, multifactorial process involving destructive remodeling of aortic wall connective tissue. Four interrelated factors involved in this process include: (1) chronic inflammation associated with neovascularization and increased proinflammatory cytokine production, (2) increased and dysregulated production of matrix-degrading proteinases, (3) destruction of structural matrix proteins, and (4) decreased medial smooth muscle cell (SMC) presence, resulting in impaired connective tissue repair. This understanding has developed from a characterization of human AAA tissue, as well as the use of different animal models that replicate human disease. The mortality of ruptured AAA is set between 40–70% in patients that manage to arrive alive in the emergency room, and that of 90% in overall patients confirmed with rAAA in the autopsy results.

A ruptured abdominal aortic aneurysm (rAAA) represents a disruption of a dilated aortic wall that leads to blood outside the aortic wall.

Aortoenteric fistula (AEF) is defined as a communication between the aorta and gastrointestinal (GI) tract. AEF is classified as primary or secondary on the basis of the underlying cause leading to the fistula development. Primary AEF is a communication between the native aorta and GI tract; secondary AEF is a communication between a reconstructed aorta (for either aneurysmal or occlusive disease) and the GI tract. The most commonly described GI tract location for primary AEF is the third and fourth portion of the duodenum (54%). It is presumed that this is due to the tethering effect of the ligament of Treitz, leaving this portion of the duodenum exposed to the direct pulsatile pressure of the aorta. Primary AEF has also been described in the following locations: esophagus (28%), small and large bowel (15%), and stomach (2%).

The first case report of PADF (primary aorto-duodenal fistula), was described by Salmon in 1843. Since then approximately 250 new cases have been reported in the literature.

Overall, the pathogenesis of primary AEF is uncertain. The proposed mechanisms are mechanical, infectious, and inflammatory. In the majority of cases, the mechanical component is
caused by the pulsatile pressure of an expanding aorta against the wall of the GI tract. This leads to local compression and ischemia, with weakening of the wall and eventual erosion with fistula formation.

Clinical presentation
The classic clinical triad for primary AEF as described by Sir Astley Cooper in 1829 consists of GI bleeding, abdominal pain, and a pulsatile mass. Recent reviews note the incidence to be 64% to 94% for GI bleeding, 32% to 48% for abdominal pain, and 17% to 25% for a pulsatile abdominal mass, with all three of these symptoms occurring concurrently in only 11% of cases. Other symptoms reported include back pain, fever, and sepsis.

In both primary and secondary AEF, a classic clinical feature is the “herald bleed.” This is a minor bleed that is self-limited because of vasospasm and thrombus formation. Herald bleeds may lead to hospital admission immediately; however, some patients experience multiple episodes of recurrent bleeding. Regardless of the initial presentation, an untreated herald bleed will commonly be followed by an exsanguinating bleed within hours to months.

Diagnosis
The diagnostic approach to the evaluation of AEF is dependent on the patient’s hemodynamic status on presentation. In general, a high level of suspicion should be maintained in approaching any patient with massive GI bleeding and a history of an aortic aneurysm or previous aortic revascularization. In such cases of massive bleeding, diagnosis will often be made during exploratory laparotomy. If the patient is stable, the three major diagnostic modalities are computed tomography (CT) with iodinated contrast enhancement, esophagogastroduodenoscopy (EGD), and angiography. There has been some controversy in the past about which study should be done first; some authors have recommended EGD, followed by CT scan, followed by visceral angiography if no source is found. However, with the improvements of radiologic imaging, CT scan has become a preferred initial diagnostic test; CT scans are less invasive than EGD or angiography, are easy to obtain, and do not risk thrombus dislodgement.

Our case report
We report the case of an 84-year-old man, who was presented in the ER on the 21st of August 2018. The patient had a two-day history of epigastric and back pain, no anemia, no history of peptic ulcer, analgesic abuse, alcohol excess, weight loss or gastrointestinal bleeding and no history of other medical comorbidities. Initial vital signs at the emergency room were stable. Five years ago he had
undergone a CABG procedure. During abdominal palpation there was no sign of regional tenderness, but the presence of a pulsating mass below the epigastric region was found. Taking into consideration the stable general condition of this patient a CT angio scan was performed showing a 6.5 x 10 cm abdominal aneurysm starting 20 mm inferior to the renal arteries that extends to the level of the iliac bifurcation. A massive anterior subintimal mural thrombus with a 30 mm thickness and no signs of leak is found at the L3 vertebral level. On the right side of the mural thrombus air bubble was detected, suggesting of an aortoenteric fistula.

![CT scan](image)

**Figures 1, 2: CT scan**

Our vascular surgery team performed an emergency operation. Right after the laparatomy, there was no evidence of retroperitoneal hematoma. A giant abdominal aneurysm could be detected after the small bowel and part of the colon were gently moved aside with the help of warm wet gauzes and retracted in order to have a clear view of the retroperitoneum. The neck of the aneurysm was located 0.8 inches inferior to the renal arteries branches. Dissection of the aortic neck and both iliac arteries was performed followed by infrarenal clampation. After opening the aneurismatic sac in a saggital plane, and after having removed the mural thrombus, a considerably 2 cm duodenal fistula appeared. The Vascular team proceeded with the aortic reconstructive intervention while simultaneously calling for help to the “on call” team of General Surgeons, in order to achieve the best possible management of the enteric fistula.
Aortic-Biiliac reconstruction with Dacron graft 16x8 and aortic wall and omentum coverage to avoid graft infection was performed. After realeasing proximal and distal clamps there was no sign of extravasation from the anastomotic sites with the graft. The hemodinamics started to normalise. The general surgeons meanwhile have arrived and a careful revision of the abdominal cavity was done, inspection of the duodenum, jejunum and ileum. Considering the impact and risks of an additional major intervention the general surgeons limited their menagement into the implantation of a duodenostomy with a T cathether (kehr). The cathether was fixed locally into the duodenal wall with prolene purse string sutures and also into the abdominal wall.

The patient made an uneventful recovery with no graft infection and no internal leak from the duodenostomy. After 17 day stay he left the hospital with the duodenostomy cathether closed and normal oral nutrition. Two months later the duodenostomy cathether was removed. Till this day the patient has been hemodinamically stable with no signs of graft infection, with no
abdominal concerns such as pain, bloating or constipation.

**Discussions**

Primary aortoduodenal fistula is very rare and challenging to manage. The importance of having a clinical or radiologic suspicion remains crucial. The short preoperative time requires a fast diagnosis based on clinical signs and Ct scan (if time allows for such a test - In cases of massive bleeding, diagnosis will often be made during exploratory laparotomy). Also the role of cell saving autotransfusion applied onto elective and ruptured AAA’s might find its benefits in PADF. Cell salvage appears to reduce overall use and exposure to allogeneic blood, and reduces length of intensive care unit and hospital stay after elective AAA repairs. There may be additional benefit by combining cell salvage with other blood-conservation techniques. Use of cell salvage in ruptured AAA repairs consistently reduced blood-product requirements.

**Conclusions**

Literature reports that PADF’s are often fatal, with a total mortality rate of 80-100% and a perioperative mortality rate of 18–63%. The diagnosis of aortoenteric fistulas are difficult because of its nonspecific and subtle clinical presentation. In cases where the main vascular vessels and the upper gastrointestinal tract are lesioned the simplest possible reconstructions come of use. Our case was managed by performing an aorto-bi iliac bypass with dacron graft 16x8 followed by the placement of a duodenostomy. The patient made an uneventful recovery with no graft infection and no internal leak from the duodenostomy and left the hospital after a 17 day stay. The guidelines that should lead into referral to the Vascular surgeon for the abdominal aortic aneurysms remains: 5,2cm in Female patients and 5,5 in Males as also in patients with an increase of >1cm per year of the aortic dilatation. Early detection of AAA’s is very important and helpful in order to improve or prevent the outcomes of patients that are presented in the ER with rAAA or PADF, and to minimise the morbidity and mortality of the disease.

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