Primary Aortoenteric Fistulae: What Every Surgeon Should Know About Its Diagnosis and Treatment

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Abstract- Aortoenteric fistula is a rare finding that is potentially fatal and is usually seen as a fistula between the aorta or an aortic graft and the third part of the duodenum. The type without the presence of an artificial graft is considered as primary and the other type as secondary. The patients usually present with the heralding symptom of minor hematemesis, which later leads to massive and mortal GI bleeding. The most important factor in the diagnosis of an aortoenteric fistula is to have a high clinical suspicion after taking an appropriate and accurate history. In hemodynamically stable patients with clinical suspicion, performing an intravenous-contrast-CT scan of the abdomen and pelvis is of assistance in diagnosis. In general, stable hemodynamics greatly reduces the suspicion for diagnosis. Treatment is often surgical, and in regard to the severity of abdominal contamination or the level of visible infection, the extra anatomical or insitu graft repair is considered as the method of choice. This article tries to transfer our experiences in two patients, each of whom presented with hematemesis and melena without a history of underlying aortic aneurysm. © 2021 Tehran University of Medical Sciences. All rights reserved.

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Introduction

The aortoenteric fistula refers to the connection between the aorta and the small intestinal tract. The fistula is formed by the erosion of both intestinal and aortic wall and a connection to each other. Aortoenteric fistulas are potentially fatal; hence untreated cases, are always mortal, and those treated surgically still carry a 30 to 40 percent mortality rate (1).

Aortoenteric fistulas are divided into primary and secondary types. The primary type is rarer and is usually caused by a connection formed between the aorta and intestine by pressure or erosive infection of the aortic wall (2). The secondary type is more common and is caused by a connection between the artificial aortic graft and the intestine. The annual prevalence of primary aortic aneurysm fistula is between 0.02 to 0.07% compared to 2.3% in patients with an artificial aortic graft (secondary type) (2,3).

AEF sufferers usually present with the heralding symptom of gastrointestinal bleeding (hematemesis and melena) with abdominal pain and occasional fever followed by recurrent gastrointestinal bleeding. Mortality is definite if left untreated or not treated promptly.

In cases with a previous history of abdominal aortic surgery, aortic fistula (secondary) diagnosis is more common in the minds of physicians, but primary fistula cases require high clinical suspicion for diagnosis, which is confirmed by CT scan with intravenous contrast. However, more than two-thirds of these cases (primary fistulas) are diagnosed only during exploratory laparotomy.

Case Report

First case

The patient was a 79-year-old man who had been treated for bladder cancer for one year and had a history

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of severe COPD. He had been consulted because of abdominal pain, melena followed by rectorrhagia. Due to low hemoglobin at 7 (g/dl) he was taken for an upper GI endoscopy, which revealed the site of bleeding as being below the second part of the duodenum.

Considering his relatively stable hemodynamics, the patient underwent an abdominal ultrasound because of abdominal pain, the result of which was an abdominal aortic aneurysm with a diameter of 37x35 mm. CT scan result was 40x44 mm diameter infrarenal aorta aneurism, which was approximately 70 mm long, containing a wall thrombosis. There was no clear fat plain between the aneurysm and the third part of the duodenum. There was as well contrast extravasation to stomach and duodenum by which AEF diagnosis came to light. Other CT findings included abdominal Ascites and bilateral Pleural effusion. His blood tests showed Cr: 1.3 WBC: 14000 and ESR: 58.

According to the above findings, the patient was immediately transferred to the operating room and underwent laparotomy, the findings of which indicated abdominal aorta aneurysm with adhesion and erosion to the third part of the duodenum where was a hematoma. Because we did not have any significant gastrointestinal fluids in the abdominal cavity and the field of operation was not extensively contaminated, the duodenum was primarily repaired, then inside of aneurysm was irrigated sterile saline. followed with by aorta endoaneurismorrhaphy by the placement of a tubular Dacron graft. The procedure was completed by inserting an omental flap between two affected sites. The patient was then transferred to the ICU, and on the fourth day after the operation, he became extubated and received chest physiotherapy. Unfortunately, two days later, he has intubated because of aspiration. Unfortunately, he died of severe pneumonia ten days later. The culture from the aortic wall was negative for bacterial infection, and histologic evaluation demonstrated only inflammation.



Figure 1. Contrast extravasation in duodenum and stomach



Figure2. Infrarenal aneurysm with the elimination of Aortodeudenal fat plane



Figure 3. Intraoperative imaging show a connection between the



Figure 4. Endoaneurismorrhaphy with tubular graft. Stitches in the upper portion of the figure show duodenal repair site

Second case

The patient was a 68-year-old man who had been referred to our Hospital due to melena and a drop in hemoglobin level. He was initially admitted by the gastrointestinal service. Due to the negative history of underlying medical disease and the lack of hematemesis and the normal appearance of NGT content at the beginning of the visit, and the stability of hemodynamics, the patient was first subjected to colonoscopy. The findings indicated that clots and black color blood were present throughout the colon. He then received intravenous fluids and was rebooked for colonoscopy as well as upper endoscopy as well to be performed within 48 hours. The next day (less than 24 hours after colonoscopy), he had massive hematemesis and had tachycardia, He was initially planned for upper emergency endoscopy, but due to his unstable condition, he transferred directly to the operating room and underwent an emergent laparotomy. The stomach and intestine were filled with blood by the look. In the surgeon's initial exploration, the aortic and iliac aneurysms were found to be about 4 cm long. The distal part of the duodenum and proximal of jejunum were severely attached to it. The vascular surgery service was asked for help. The operation went through Aorta and Iliac explore, and after meticulous proximal and distal control, the small intestine was freed from aortic adhesion, the result of which revealed a 2 cm diameter fistula between the aorta and D4. The intestine was initially repaired in two layers. In the absence of abdominal cavity contamination, endoaneurismorrhaphy was performed, and reconstruction was completed by the Aortobyiliac bypass technique. An omental patch was placed and fixed between the Aorta affected site and duodenum. Post-operation, the intubated patient was transferred to the ICU, and blood pressure was dependent on norepinephrine, and the patient's condition did not

improve. Unfortunately, he died the day after the operation. The culture from the aortic wall was negative for bacterial infection, and histologic evaluation demonstrated only inflammation.

Discussion

Aortic enteric fistulas are very uncommon, but due to the mortality rate of nearly 100% if left undiagnosed. So, it is important to have a clinical suspicion for an early diagnosis. It is important to have a history of previous illnesses and surgeries, and most patients are not usually referred to a vascular surgeon in the first place if so only after upper endoscopy and failure to observe the source of GI bleeding or when the source of bleeding found to be in the distal to D2.

Aortoenteric fistula was first described by Cooper in 1822. The cause of the primary aortic aneurysm is not well known, unlike the secondary type 4. There is a strong theory about it, and it is that erosion occurs between a part of the duodenum and aorta. The erosion is aggravated by infection, inflammation, or foreign bodies. The involved part of the duodenal site is usually attached to the retroperitoneum and passing anterior to the aorta (4,5).

In a report, God *et al.*, found out that 88% of primary cases of abdominal aortic aneurysms were caused by atherosclerotic aneurysms. However, primary fistulas have also been reported in traumatic or mycotic aneurysms, and to a lesser extent due to cases such as radiotherapy, pancreatic carcinoma, gallstones, diverticulitis, appendicitis, and media cystic necrosis (6,7,8).

The cause of most cases of aortic aneurysms is attributed to the high levels of abdominal aortic wall metalloproteinase, which can cause inflammatory proteolytic degeneration, along with the specific previously discussed anatomical condition of the duodenum, which can cause primary fistula (9).

In Primary fistulas reported, the most common site of involvement is in the duodenum (83%), most of which involve the third part (57%). Other affected areas include the other parts of the duodenum (17%) and the fourth part of the duodenum (6%) (10).

Aortic aneurysms usually present with signs and symptoms of upper gastrointestinal bleeding (64%), pain (32%), and abdominal pulsatile mass (25%) (11). It should be considered in all patients with a previous history of abdominal or thoracic aortic aneurysm or a history of aortic surgery with an artificial graft and should undergo appropriate diagnostic tests immediately, although it is clear that diagnostic investigations should not be prioritized in the price of delaying immediate surgery if the patient's hemodynamics are unstable.

The time between the herald GI bleeding and the onset of fatal bleeding varies, and it is reported that more than 70% of patients have at least 6 hours to initiate massive bleeding (12).

As previously recommended, the patient should first undergo upper endoscopy and then a CT scan, and if the bleeding source is still not found, angiography will be the next. However, with the advancement of equipment and the improvement of CT scan quality, it is currently recommended that the patient undergo a CT scan with intravenous contrast material in case of clinical suspicion of aortoenteric fistula because it is non-invasive and can be performed promptly without the risk of clot mobilisation12. The disappearance of the aneurysm wall of the fat plane between the aorta and the duodenum, or the presence of air in the retroperitoneum, are major radiologic signs of an aortic aneurysmal fistula.

Upper endoscopy is not very sensitive and specific for the diagnosis of aorticenteric fistula and may require several repetitions. Its diagnosis is strongly operator related and therefore is more helpful in excluding other causes of acute upper gastrointestinal bleeding (12,13).

The treatment for primary aortic aneurysm fistula is not clear due to its very low prevalence; however, surgery is still considered to be the main treatment. The selection of the type of surgery is based on the cause of fistula, level of infection, and retroperitoneal contamination and is aimed to prevent fistula formation in the future.

In the case of primary fistulas in mycotic aneurysms, extensive debridement of infectious tissues and extra anatomical reconstruction of the aorta is recommended (14). However, if the rate of intra-abdominal contamination is low, in-situ grafting may seem more reasonable (15).

If the tissue or fluids lab culture turned to be positive, the patient should be treated with antibiotics for 4 to 6 weeks after the operation based on the culture and antibiogram. If culture is negative, the antibiotic should be given for 7 to 10 days empirically (16).

Primary aortic aneurysms have a worse prognosis in regard to infectious aortic aneurysms than other cases, and their mortality rate after surgery is above 50%. Perhaps the reason for it is the length of operation time for its extra anatomical reconstruction.

In cases where there is no evidence of infection, the use of an in-situ aortic graft and intestinal repair and the insertion of an omental flap between the aorta and intestine is necessary and sufficient (17).

In the end, it is mentioned again that the most important principle in the survival of patients with aorticenteric fistula is their early diagnosis and prompt treatment, which is achievable by obtaining a complete and accurate history and physical examination, high clinical suspicion, and prompt Abdominal and pelvic CT scans.

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