



Aorto-Enteric Primary Fistula on Remote Endovascular Aneurysm Repair

CASE REPORT

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ABSTRACT

Aorto-Enteric Fistula (AEF) can be primary or secondary to surgery. The authors describe a case of death due to acute gastrointestinal hemorrhaging, resulting in primary AEF, in a patient who had undergone surgery in the past. The site and the pathological features of the lesion allow us to recognize the primary nature of the AEF, excluding remote surgery as a possible cause. The differential diagnosis is based on the patient's history and also on the anatomical characteristics of the lesion, such as location and extent.

Keywords: Aorto-enteric fistula, death, endovascular aneurysm repair

INTRODUCTION

Aorto-Enteric Fistula (AEF) is defined as a direct communication between the aorta and the intestinal lumen (1). There are two forms of AEF when surgical procedures are performed in the abdominal aorta (2): primary aorto-enteric fistula (PAEF) or secondary to Endovascular Aneurysm Repair (EVAR). The secondary form is much more common (3). The incidence rate of the secondary form is 0.36%–1.6% versus 0.04%–0.07% for PAEF (4).

There are two theories about the pathogenesis of AEF: (i) repeated mechanical trauma between the pulsating aorta and duodenum, which causes fistula formation, or (ii) a low-grade infection could be the primary event, which causes abscess formation and subsequent erosion through the bowel wall (5).

The prevention is the treatment of hypertension and to provide adequate surgery and monitoring, post-surgery. Diagnosis of AEF requires signs of infection and of gastrointestinal bleeding and is determined by the use of esophagogastroduodenoscopy and computed tomography (4).

Urgent surgery is still the recommended treatment (6); other possible options are in situ aortic reconstructions using prosthetic grafts or axillo-bifemoral bypass (7). In this sense, the literature identifies PAEF as a rare complication of aortic aneurysm and a rare cause of gastrointestinal bleeding which can result in death (8).

Since 1817, there have only been about 300 reported cases of PAEF in the (English) literature, according to Lee et al. (9), and about 350 to 2006 according to other authors (10-12).

We describe a case of death, resulting from gastrointestinal bleeding, due to aorto-duodenal fistula (third part of duodenum) in a patient who, in the past, had been treated by surgery.

CASE REPORT

A 74-year-old male with a history of hypertension, dyslipidemia, and who had undergone open surgical intervention (15 years earlier) for aortic endovascular prosthesis (iliac excluded). Subjected to normal follow-up echo, for an indefinite amount of time (there are no recent echo or CT), he had no symptoms in the following history, as reported by family members.

The man was found in his home, lying on the ground, in a pool of blood. As determined via external examination, there were no signs of violence; the only finding was a xipho-pubic scar. The subject measured 170 cm in height, and his weight was approximately 75 kg. Examination of thanatological phenomena showed a small degree of hypostasis, which showed as being pinkish and was positioned on the dorsal surface of the body. There were no signs of rigidity in the joints and there was an absence of putrefaction.

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Figure 1. Aortic bisiliac prosthetic

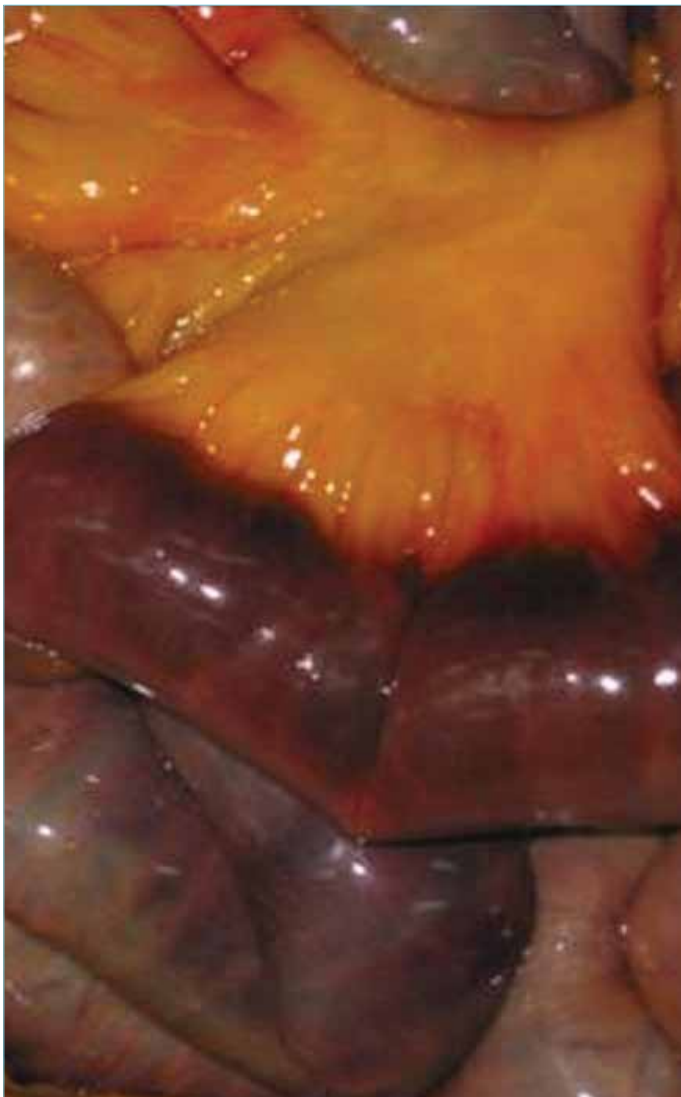


Figure 2. Appearance of the intestinal loops

The heart had increased in size and volume (420 g in weight, longitudinal diameter was 12.5 cm, transverse diameter was 12.5 cm, and the anteroposterior diameter was 4 cm). The thickness of the free wall of the left ventricle was approximately 1.2 cm, whereas it was 1.1 cm at the level of the septal. In the longitudinal section, some focal areas of widespread myocardiosclerosis were noted, but the integrity of papillary muscles and chordae tendineae was maintained. Calcified atheroma and stenotic plaques were found in the coronary lumen.

The lungs had increased in size and changed in texture (right lung was 850 g; left lung was 650 g) and sputtering pressure. Large, medium, and small bronchi were found with patent lumen and hyperemic mucosa. The parenchyma section appeared congested, and, when squeezed, a foamy substance was found. During the examination of the abdominal cavity, the aortic bisiliac prosthesis (Figure 1) was noticed immediately, and a normal consistency of appearance for the stomach and intestinal tract was observed but were dilated and had dark red walls (Figure 2).

Diffuse atherosclerotic plaques were found in the section on the longitudinal plane of the thoracic aorta. In the abdominal portion of the aorta, however, an aneurysmal dilatation (6-cm-long) was found at the high site (at the level of the third lumbar vertebra), with a widely thrombosed lumen and communicating via duct fistula with the rear wall of the overlying duodenum (Figure 3). At the time of histology, diffuse atherosclerotic plaques were confirmed, complicated with endoluminal thrombosed material and thinning of the wall adherent to the duodenum at the level of the thoracic and dissecting aneurysm of the abdominal aorta.

DISCUSSION

In the case of AEF that is secondary to surgery, the most common cause is endoleak, or persistent bleeding, with a volumetric increase of the aneurysmal sac, favored by dislocation or erosion of the aortic graft, or tissue degeneration resulting from fixation hooks or metallic stents. More generally, any cause of inflammatory aneurysm can cause AEF (13).

The hypothesis of an AEF secondary to inflammatory aortitis due to surgery (14) is unlikely; the time needed for the development of AEF is variable, the surgical treatment was remote relative to the time scales described in the literature for fistula (formed in the vicinity of the intervention) (1). The site of the aneurysm and fistula was different than that of surgery (15). Thus, it was a primary form.

In PAEF, undoubtedly, the most frequent cause is atherosclerosis, especially if, as in this case, other locations of the aorta and the heart prove to be affected by atherosclerotic disease. In the literature, it is confirmed that there is an association between atherosclerotic heart disease and aortic aneurysm (16, 17). Atherosclerosis is associated with fistula, even in the absence of aneurysmal dilatation, and the most frequent sites are the third and fourth portions of the duodenum, due to their proximity to the aorta (18, 19). Other causes, such as septic aortitis, tuberculosis, cancer, radiation, and foreign bodies, are less frequent (12).

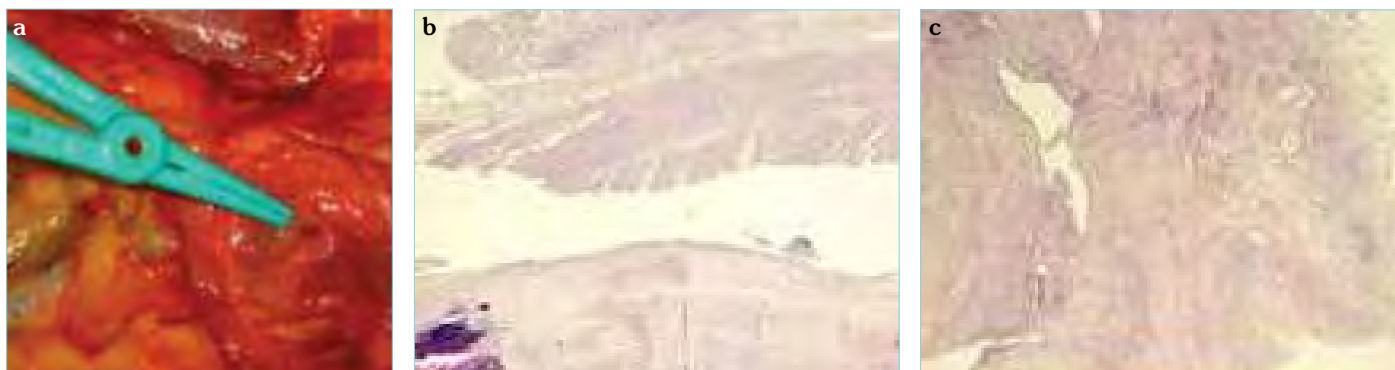


Figure 3. a-c. Macroscopic (a) and microscopic details in longitudinal (b) and transverse (c) sections of the aorto-duodenal fistula

The case subject did not have intermittent herald bleeding, which is a usual manifestation of AEF (20), likely due to self-limitation by the thrombus, determined as aneurysmal lumen or spasm of the intestinal wall around the fistula (12, 21).

Pulsating aortic lead ischemia and subsequent necrosis of the intestinal wall, causing the subsequent formation of a fistulous communication of aorta with the intestine and rapid exsanguination, were found (22).

The diagnosis was complicated by the fact that the acute symptoms are superimposable to the more-common duodenal ulcer (23). However, an aortoduodenal fistula is rarely appreciable using endoscopic examination, and, in case of the presence of gastritis or ulcers, cannot exclude the presence of a fistula (24, 25).

Unfortunately, cases of subacute or intermittent bleeding, which leave the time–depth diagnosis are less frequent (1). Mortality, 100% in the case of massive hemorrhage which is not followed by surgery, is high, even when treated surgically (30%–40%) (19). Thus, the subject might have been saved if he had been quickly treated with surgery. The massive loss of blood caused, as in this case, ischemia of the main organs and systems, resulted in rapidly evolving acute heart failure.

CONCLUSION

The case patient's history can only generate suspicion of AEF. Esophagogastroduodenoscopy or CT are useful for diagnosis, but in cases of death the anatomical characteristics of fatal injury, such as location and the time of the onset of bleeding can be more important.

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case: CB., PG., AC. Performed the experiments or case: FM., SR. Analyzed the data: PG., CB. Wrote the paper: PG., FM. All authors have read and approved the final manuscript.

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