FÍSTULA AORTO-ENTÉRICA, ESTADO ATUAL DA ARTE

AORTOENTERIC FISTULA, CURRENT STATE OF THE ART

Inês Antunes*, Carlos Pereira¹, Carlos Veterano¹, Gabriela Teixeira¹, Carlos Veiga¹, Daniel Mendes¹, Henrique Rocha¹, João Castro¹, Rui Almeida¹

1. Serviço de Angiologia e Cirurgia Vascular, Centro Hospitalar do Porto, Porto, Portugal

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RESUMO

A fístula aortoentérica (FAE) representa uma causa rara da hemorragia gastrointestinal. Existem dois tipos de FAE: as primárias e as secundárias. As FAE primárias ocorrem normalmente em associação a aneurismas da aorta abdominal. As FAE secundárias estão associadas à presença de material protético em relação com cirurgia aórtica prévia, normalmente associados a infecção da prótese vascular e representam o tipo mais comum de FAE. Um alto nível de suspeição é fundamental para o diagnóstico. Se não prontamente diagnosticada e tratada, a taxa de mortalidade é muito alta. O papel do tratamento endovascular nestes casos ainda não está definido. O nosso objetivo é fazer uma revisão não sistemática da literatura disponível relativamente à etiologia, apresentação clínica, diagnóstico e tratamento das FAE.

Palavras-chave
Fístula aorto-entérica; Aneurisma aorta abdominal; Aneurisma artéria ilíaca; Hemorragia gastrointestinal; Dor abdominal

ABSTRACT

Aortoenteric fistula (AEF) is a rare cause of gastrointestinal (GI) bleeding. There are two types of AEF: primary and secondary. Primary AEF usually occurs in association with abdominal aortic aneurysm (AAA). Secondary AEF are associated to aortic grafts, normally in relation to graft infection, and represent the most common type. A high level of suspicion is essential to a prompt diagnosis. If not promptly diagnosed and treated the associated mortality is very high. The role of endovascular treatment is not yet defined. Our aim is to perform a non-systematic review of the available literature concerning etiology, clinical presentation, diagnosis and treatment of AEF.

Keywords
Aortoenteric fistula; Abdominal aortic aneurysm; Iliac artery aneurysm; Gastrointestinal bleeding; Abdominal pain

ARTIGO DE REVISÃO

INTRODUÇÃO

Aortoenteric fistulae (AEF) is a communication between the aorta and the gastrointestinal tract. It is a rare but life-threatening cause of gastrointestinal (GI) bleeding¹. AEF can be primary or secondary². Primary AEF arise from the native aorta and normally are related to abdominal aortic aneurysms that erodes the bowel wall and rupture into the enteric lumen. The duodenum is the most common site of communication²⁻³. Secondary AEF are associated to aortic grafts, normally in relation to graft infection, and represent the most common type. A high level of suspicion is essential to a prompt diagnosis. If left untreated, the mortality rate of AEF is about 100% and even with treatment the mortality is high. Conventional surgery remains the standard treatment, the role of endovascular treatment is not yet defined. The aim of this paper is to perform a non-systematic review of the available literature concerning etiology, clinical presentation, diagnosis and treatment of AEF.

ETIOLOGIA

AEF can be primary or secondary. Primary AEF arise from the native aorta and normally are related to abdominal aortic aneurysms that erodes the bowel wall and rupture into the enteric lumen.
The duodenum is the most common site of communication (2–5) usually the third portion (83%), but the jejunum (4%), sigmoid colon (4%), stomach (4%) and the ileum (2%) may also be involved (6). The etiology of AEF has not been fully understood. Most authors believe that inflammatory destruction plays a major role. In fact, studies of the abdominal aortic aneurysms wall detected an increase in metalloproteinases (8) which leads to inflammatory cell infiltration and eventually inflammatory cell degeneration. Most authors believe that due to the proximity of the aorta and the duodenum associated with its fixed nature, the growing of the aneurysm can cause local inflammation which could lead to a fistula. Although 80% of primary AEF involve aortic aneurysms, they can be associated to other causes such as GI ulcers and foreign bodies, irradiation, inflammatory conditions such as diverticulitis, malignant tumors and iatrogenesis (7).

Secondary AEF occurs in association with aortic grafts in patients with previous aortic surgery and represent the most common type. The mechanism underlying the development of a secondary AEF remains unknown but there are two postulated hypotheses: the first one argues a relation to prosthesis infection and inflammatory response to this infection and the other suggests a physical stimulation (8). The last hypothesis defends that continuous stimulation due to aortic pulsation transmitted by the graft directly affect the bowel wall and is supported by the fact that most secondary AEF involve the third or fourth duodenum portion. This hypothesis points out the importance of avoiding contact and consequent adhesion between graft anastomosis and the intestinal wall by interposing native tissue between them in primary surgery.

**CLINICAL PRESENTATION**

The classical clinical symptom triad, but only present in 11% of patients (9), consists in gastrointestinal bleeding, abdominal pain and pulsatile abdominal mass (10). Hemorrhage is the most common sign, but it varies from intermittent to massive life-threatening bleeding. Most patients have a history of intermittent self-limited hematochezia or hematemesis before the episode of massive bleeding. These herald bleedings can precede massive bleeding hours to a month and in one third of massive bleeding caused by AEF occurs within the 6 hours after the herald bleeding (11). The mechanism adjacent to these intermittent bleedings is thought to be related to vasospasm and thrombus formation due to transitory decrease in blood pressure and a temporary decrease in blood flow in the fistula, which allows hemostasis. Restoration of normotensive pressure causes displacement of the thrombus, leading to another bleeding. Septicemia and fever may be present (12) and usually are related to advanced condition.

**DIAGNOSIS**

AEF should be suspected in all patients with gastrointestinal bleeding and a history of an aortic aneurysm or a previous aortic revascularization particularly with a graft. As the initial presentation could be self-limited bleeding, usually hematochezia, the diagnosis should be made in this time period, before massive bleeding. The best diagnostic modality is not yet defined. In the past, some authors recommended endoscopy as the first line diagnostic tool, if negative a CT scan should be performed and, if negative, the patient should be submitted to angiography (12,13).

In the last years, there has been a great improvement in the angio CT scan and it has become the preferred initial diagnostic tool. Nowadays angio CT scan has a reported sensitivity of 94% and specificity of 85% (14) in the diagnose of AEF and also has the advantage, in comparison to endoscopy and angiography, that has no risk of thrombus dislodgement and trigger hemorrhage. Angio CT findings include: air around the aorta/aortic graft, loss of the fat plane between aorta or the aortic graft and gastrointestinal tract, bowel wall edema around the aorta graft and the presence of contrast material into the gastrointestinal tract.

Despite the advances it remains a difficult diagnosis. Because of the occurrence of intermittent bleeding, if the diagnostic exam was performed in the period of hemostasis, it can’t be unable to diagnose the source of bleeding and for that reason, when there is a high suspicion even with a normal angio CT scan, angiography or endoscopy, the diagnosis of AEF cannot be excluded.

**TREATMENT**

If left untreated, the mortality rate of AEF is about 100%. Even with treatment the mortality is high (estimated to be between 30–40%).

AEF treatment should take in account etiology and degree of abdominal contamination and should include bleeding control, repair of the bowel defect, restore circulation and eradicate infection (15).

Regarding primary AEF, surgical repair of AAA and of the bowel defect is the standard treatment. When there is no imagiological/intraoperative evidence of infection, an anatomic repair with a graft associated with bowel defect repair may be performed. It is important to cover the graft with an omentum flap, in order to prevent a future secondary AEF based on the physical stimulation theory. If there is gross evidence of infection, aneurysm repair associated...
with extensive retroperitoneal debridement with omentoplasty and an extra-anatomic bypass should be performed. In comparison to in situ reconstructions, this procedure has higher rates of mortality and limb loss and for that reason is normally only used when there is evidence of retroperitoneal purulence or exuberant inflammation.

All patients with AEF should receive broad-spectrum antibiotics in the postoperative period that should be maintained until culture results are available. In the case of negative results broad-spectrum antibiotics can be discontinued after one week. If the cultures are positive a tailored antibiotic should be maintained for 4 to 6 weeks.

Concerning secondary AEF with evidence of graft infection, it is an agreement that all infected graft material should be resected and arterial tissue should be debrided until arterial tissue capable to support an arterial suture was found. The defect in the gastrointestinal tract should be repaired. An omentum flap should be interposed between arterial suture and the bowel. After that, there are many options concerning limb revascularization. When the previous graft has been placed due to occlusive disease or has a previous reported occlusion, it may be possible not to proceed with lower limb revascularization strategies. When reconstruction is necessary extra-anatomic bypass or in situ reconstruction could be used depending on the degree of intra-abdominal contamination. In the presence of evident retroperitonitis, retroperitonel debridement and drainage must be considered and an extra-anatomic bypass should be performed. In situ reconstructions may be associated with some benefits such as a possible decreased risk of aortic stump blowout and a decreased risk of renal arteries thrombosis. In situ reconstruction may be attempted using cryopreserved allografts, autogenous conduit (neo-aortoiliac) or grafts (namely antibiotic-soaked or silver-coated grafts). Chung et al. advocated that the neo-aortoiliac procedure with the use of femoropopliteal vein as conduit combines the benefits of lowering the risk of aortic stump blowout and avoids the risk of infection inherent to the use of graft material. Regarding the use of antibiotic-coated grafts it may be an option, in fact in 2006 O’Connor et al published a meta-analysis concluding that in situ reconstruction with rifampicin-bonded grafts had lower mortality in comparison to extra-anatomic reconstructions. Antibiotic therapy also has an important role in the management of secondary AEF. Empiric antibiotics to cover the most common microorganisms should be started immediately and intraoperative cultures should always be obtained. If positive cultures were obtained antibiotics must be directed to isolated microorganisms for a period between 4 to 6 weeks.

The role of endovascular repair has not yet defined. Endovascular repair is a tempting option, in part because of the simplicity of the treatment, but there is no direct observation of the abdominal cavity and it does not allow the repair of the bowel defect. Also, as it evolves a deployment of a graft in a potentially infected territory, there is a great concern about future graft infection. In recent years, endovascular repair has been used as a treatment option in elderly patients, or patients too frail for conventional surgery.

For secondary AEF, endovascular treatment emerged for haemorrhagic control, as a bridge for more definitive repair. Endovascular treatment can represent a more definitive solution in primary AEF, since there is no graft material to be removed and normally the associated infection is easier to control. In 2009 a systematic review on this issue was published including 41 patients with AEF that were initially treated with EVAR. Successful endograft implantation was obtained in all patients. In 20% a concomitant bowel repair was performed. At 13 month of follow up 44% of the patients developed infection or haemorrhage and authors concluded that EVAR should be considered only as a bridging procedure until definitive treatment can be performed. In 2011 a multicentre retrospective study involving 25 patients with AEF reported a short term benefit for EVAR in comparison to open repair (overall morbidity 25% for EVAR vs 77% for open repair) but this initial benefit was lost by the second postoperative year due to an increase in recurrence of infection, bleeding and AEF.

DISCUSSION/CONCLUSIONS

AEF is a rare differential diagnosis of lower GI bleeding. Despite being a fatal condition when not treated, there is still poor awareness that gastrointestinal bleeding can be caused by AEF. The diagnosis is difficult, and it is apparent that complementary study (with angio CT scan, endoscopy or angiography) may not show the source of haemorrhage if performed during the haemostasis period with clot formation after an herald bleeding. Thus, if there is a strong diagnostic suspicion, this cannot be excluded due to the non-demonstration of the source of haemorrhage in the complementary study. Until now, conventional surgery is the standard treatment, but the best treatment is not yet defined. Endovascular treatment represents an alternative for patients who are poor candidates for surgical treatment or as a bridge for definitive treatment in the case of secondary AEF but can represent a more definitive treatment for primary AEF depending on the degree of contamination, aneurysm type and individual characteristics of the patient.
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