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A PATIENT WITH THREE AORTOENTERIC FISTULAS IN A PERIOD OF FIVE YEARS: CASE REPORT

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ABSTRACT

Background: Aortoenteric fistula (AEF) is a pathological communication between the aorta and gastrointestinal tract that presents a life-threatening condition. It can be primary or secondary, based on the underlying cause of fistula development.

Case study: We present a 67-year-old female patient who suffered from three secondary AEFs in a period of five years. After two abdominal surgeries for gastric ulcer and colorectal adenocarcinoma (TNM stage II), the patient had an open abdominal aortic aneurysm reconstruction. For each AEF presentation, opened surgical reconstruction was performed.

Conclusion: Morbidity and mortality rates after AEF surgery are high despite advances in surgical techniques and materials. Three times recurrent AEF in a single patient with 5-years survival after initial reconstructive surgery is rare event.

Keywords:

Aortoenteric fistula, aortic surgery (AEF), aortic reconstruction complication, abdominal aortic aneurysm (AAA)

INTRODUCTION

Aortoenteric fistula (AEF) is a pathological communication between the aorta and gastrointestinal tract. It can be primary or secondary, based on the underlying cause of fistula development [1]. A primary AEF occurs naturally, without any prior aortic reconstruction. It can develop due to compression of the AAA on the duodenum, local inflammation or aortic infection (peptic ulcer perforation into the aorta or tumor erosion of the aorta) [1].

A secondary AEF forms following some surgical intervention such as an aortic reconstruction. The AEF usually affects the distal third or fourth region of the duodenum, but can also affect the proximal jejunum [2]. In the majority of cases, the initial event leading to this condition is a prosthesis-infection, leading to a weakening of the suture line. This subsequently leads to a false aneurysm and AEF formation [3]. In a small number of cases continuous pulsatile pressure of the aortic graft to duodenal compression or duodenal injury leads to AEF formation [4].

Diagnostic options in detecting this condition include esophagogastroduodenoscopy (EGDS), multislice computed tomographic angiography (MSCTA) and scintigraphy with radiographically marked red blood cells: all of which are seldom positive (MSCTA in 11%) [5,6]. Due to these low rates of true positive findings, clinical examination and physicians' opinion remains the most important diagnostic tool.

AEF presents a rare and particularly complicated problem in vascular surgery. When left untreated, the outcome is usually fatal. Surgical repair is fraught with complications, morbidity and mortality rates remain high despite advances in surgical techniques and materials [1].

A recurrence of AEF following surgery in the same patient is an extremely rare clinical finding.

CASE STUDY

A 67-years old female patient came to the emergency department because he collapsed and had signs of melena. Her blood pressure initially was 95/70 mm Hg, heart rate 90 bpm and hemoglobin concentration of 65 g/l. She had a history of stomach ulcer surgery, right hemicolectomy due to colorectal adenocarcinoma (TNM stage II) and chronic renal failure. Also, surgical reconstruction of the abdominal aorta using a Dacron graft (Vascutec Gelsoft Plus, Terumo, Tokyo, Japan) due to an abdominal aortic aneurysm (AAA) was performed in another institution six years prior to admission. An emergency EGDS was performed which did not show any active or recent bleeding. There were no signs of a visible aortic graft through the bowel or other signs of bowel perforation. MSCTA showed periaortic hematoma and active extravasation from the aorta directly into the bowel which was adherent to the previously reconstructed aorta – an AEF (Figure 1). Emergency surgery was performed during which the rupture of the proximal and distal aortic anastomosis was discovered and an AEF with the proximal jejunum. Due to the patient's hemodynamic instability, only re-suturing of the proximal and distal aortic anastomosis of the existing aortic graft was performed. The bowel perforation was directly sutured in two layers.

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Following surgery, the first twelve postoperative days went uneventful. On the 13th day the patient had recurrent stomach pain and hematochezia, with consequent blood loss resulting in a hemoglobin count of 68 g/L . A MSCTA was ordered which showed the possibility of active extravasation in the previously operated area. The patient underwent to a second surgical procedure during which active bleeding and anastomotic disruption was ruled out.

After six weeks of antibiotic treatment with teicoplanin, meropenem, imipenem, cilastatin and fluconazole, the patient was discharged.

That same year the patient was re-operated in another hospital due to a new AEF formation. During that procedure the old prosthesis was removed and replaced with a new Dacron prosthesis (Interguard Silver, Maquet, Rastatt, Germany).

Four years after the second procedure due to AEF, the patient presented with hematemesis and hematochezia (lasting one day) in the emergency department. An MSCTA revealed a newly formed AEF with active bleeding into the bowel. Surgery was indicated and the patient lost consciousness just before entering the operating theatre. Her condition of hypovolemic shock was accompanied with decreased blood pressure (BP 70/40 mm/Hg), tachycardia (HR 123 bpm) and consequent cardiac arrest. Cardiopulmonary resuscitation was initiated and a heart rhythm was restored. During the surgery, removal of the previous graft was performed and a new aortic reconstruction (Silver Graft, B. Braun, Berlin, Germany) and bowel suturing were completed successfully. Despite all efforts, the patient died two hours after surgery in the intensive care unit due to cardiopulmonary failure.

DISCUSSION

AEFs are rare, but when they present are very severe and urgent conditions, usually demanding prompt diagnostics and treatment. The diagnostic capabilities are not very specific and it is very difficult to diagnose an AEF with any imaging methods available or with EGDS. It is therefore usually left to clinical suspicion and physician experience that guides the surgeon towards the diagnosis and indication for emergency surgical treatment. Secondary AEFs are seen much more frequently than the primary cases [7]. In the case of a secondary AEF, diagnosis is often easier because it is usually seen in a patient who previously underwent an aortic reconstruction. These patients commonly present with melena and "herald bleeding".

However, recurring AEF's following previous surgical reconstruction is a rare occurrence. The patient we presented in this paper presented with her first secondary AEF one year after the initial reconstruction of the AAA. Nevertheless, she later presented with two more AEFs which resulted in three AEF surgeries. She survived five years after the initial AEF reconstruction which is an excellent result.

There are many methods of AEF reconstruction such as local graft repair, excision of the graft without the reconstruction, in situ graft replacement and extra-anatomic revascularization with graft excision or delayed excision after two to three days. All the mentioned methods are accompanied with a high mortality rate and it is still doubtful which is the best choice [8]. In this patient, several methods were performed.

The first surgery included re-suturing the old prosthesis. This method was chosen due to the poor hemodynamic instability of the patient. Although this method has been associated with worst outcomes, high rates of reinfections and high mortality rates, in this situation, this was the only possible option to save the patient [9-12].

The second AEF surgery was done in another hospital almost one year after the first. In this procedure the old graft was removed and replaced with a new prosthesis.

Five years after the initial AEF reconstruction surgery, the patient developed a third AEF. At this point the patient was in a very poor clinical condition. Despite the difficulty to operate this patient due to the previous abdominal surgeries, the reconstruction was successful and a new graft was placed. The patient died couple hours following surgery.

CONCLUSIONS

AEF presents as a rare and extremely complicated condition in vascular surgery. There are vast arrays of surgical procedures available which can lead to many outcomes depending on the patient. Using the appropriate surgery in critically ill patients is imperative and it is the goal of the surgeon to give them the best chance of survival,

Unfortunately, after multiple efforts our patient eventually died as an indirect result of her AEF. This patient was our first to have survived two previous AEF surgeries and the only patient who had developed a total of three in her lifetime.

CONFLICT OF INTEREST:

The authors declare that there is no conflict of interest.

The patient gave her informed consent prior to her inclusion in case report.

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FIGURES

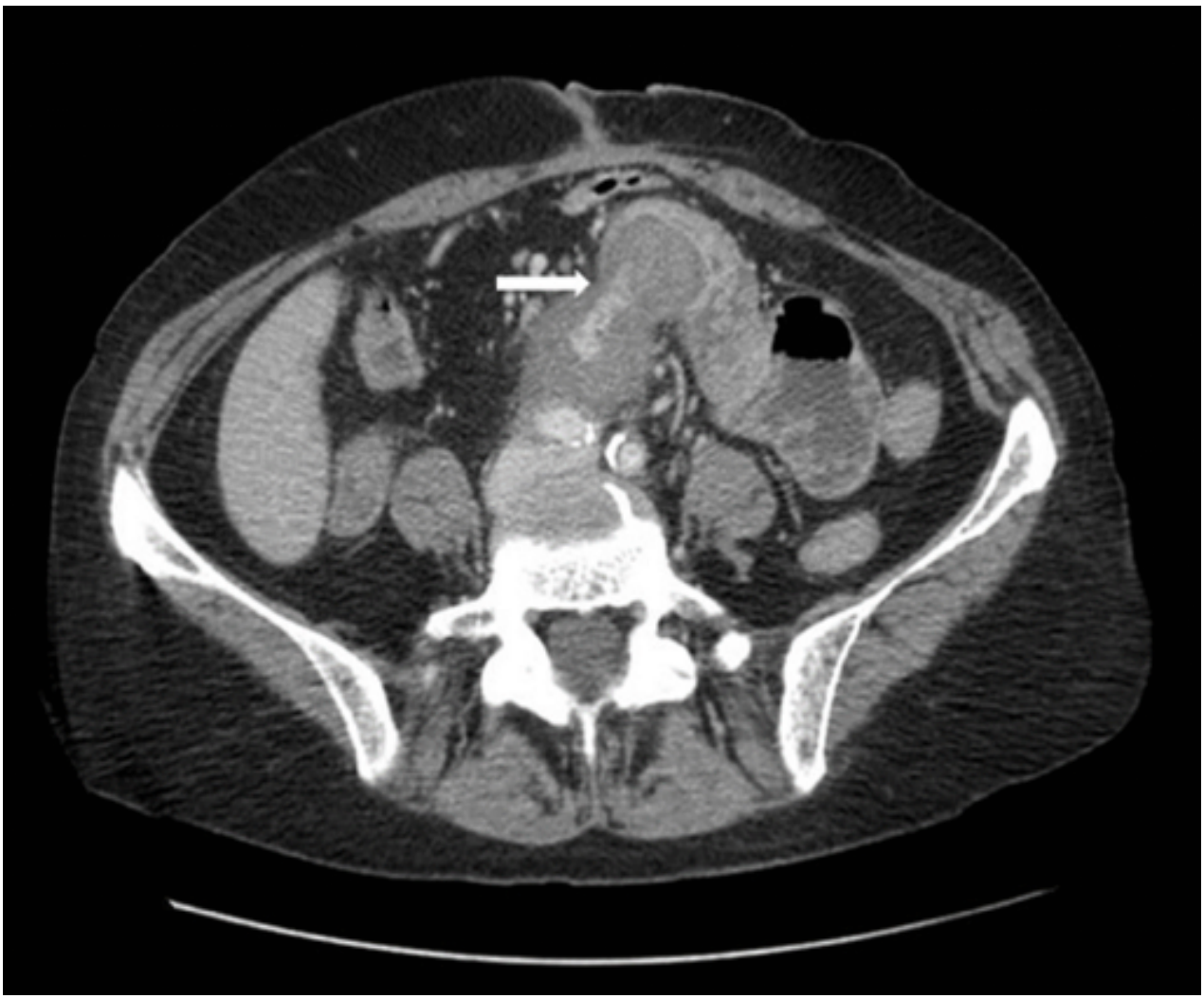

Figure 1. Arrow in the image shows the position of aortoenteric fistula (AEF)