

Aortoenteric fistulas – our experiences with surgeons' nightmare

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Background: Aortoenteric fistula (AEF) is a rare medical emergency, but one of the most difficult and threatening complications of gastrointestinal (GI) bleeding.

Methods: A retrospective observational study was performed on patients hospitalised with GI bleeding and a definitive AEF diagnosis. We collected operative reports and medical records of patients operated on with an AEF diagnosis. The literature data and our data were analysed and discussed.

Results: We admitted eight patients who were definitively diagnosed with AEF after reviewing our hospital records. All patients were male except one. Their ages ranged from 28 to 82, with a mean of 64. All but two patients had secondary AEF (SAEF). Four SAEF cases had open aortic surgery and three had a history of endovascular procedure. The main complaints of the patients on admission were poor general condition, abdominal pain, and GI bleeding. Melena was found in all patients. Hematemesis and hematochezia were other significant GI bleeding findings. Infected grafts were removed in all but one patient; extra-anatomical bypass surgery and bowel repairs were performed. One patient underwent endovascular repair. In all patients, the 30-day in-hospital mortality rate was 50%.

Conclusion: In patients presenting with GI bleeding, an aortoenteric fistula should be considered. The outcome depends on early diagnosis, the patient's medical status, the severity of infection, and the anatomic location of the affected aorta. A multidisciplinary approach, appropriate treatment planning and close follow-up after treatment lead to positive outcomes.

Keywords: aortoenteric fistula, diagnosis, gastrointestinal bleeding, surgical treatment

Introduction

Aortoenteric fistulas (AEFs) are a medical emergency and one of the rare causes of fatal gastrointestinal (GI) bleeding, often occurring after aortic aneurysm surgery and rarely as a result of primary aortic or intestinal pathologies.¹⁻³ While most AEFs are regarded as a secondary complication following prior aortic surgery, they seldom develop primarily. The location of the fistula usually occurs between the abdominal aorta and the duodenum.⁴

Early diagnosis and surgery are required to manage AEF. Unfortunately, these types of cases often have intermittent and nonspecific findings, making diagnosis difficult.^{5,6} Diagnostic delay can worsen prognosis, especially in unstable septic patients with severe comorbidities. This situation simulates a nightmare for the clinicians and surgeons who are responsible for the care of the patient. The promptness of the diagnosis, the medical condition of the patient, the presence of severe infection and the anatomical site involved all affect the outcome.⁷ Despite all this, the use of planned up-to-date diagnostic and treatment methods and a multidisciplinary approach will reduce morbidity and mortality in AEF cases.

In this paper, we aimed to present our experiences in diagnosing and treating AEF patients in our clinic from 2010 to 2020, together with an analysis of the literature.

Materials and methods

A retrospective observational study was conducted of patients hospitalised with GI bleeding and a definitive diagnosis

of AEF between 2010 and 2020 in the cardiovascular surgery department of Karadeniz Technical University. The local ethics committee approved the study protocol and stated that the study did not fall under the Medical Research Involving Human Subjects Act and, therefore, an informed consent form was not required.

We collected the operative and medical records of patients who underwent surgery with a diagnosis of AEF in the cardiovascular surgery department. Data were analysed, including demographic information, clinical manifestations and complications, physical examinations, laboratory and imaging findings, treatments and outcomes. The diagnosis of AEF was confirmed by endoscopy, radiographs and/or surgical findings. In addition, we evaluated the literature data and our data by scanning the literature.

Results

We enrolled eight patients who had a definitive diagnosis of AEF after reviewing our hospital records between 2010 and 2020. All patients were male except one. Their ages ranged from 28 to 82, with a mean of 64. All but two of the patients had secondary AEF (SAEF). In the medical history of patients with SAEF, four had open aortic surgery and two had endovascular procedures. Four SAEF cases had open aortic surgery and two had endovascular procedure in their medical histories. A patient diagnosed with primary AEF (PAEF) had a clinical history of gynaecologic surgery and radiation therapy. The fistula point in this patient was at the junction of the proximal right iliac artery and the proximal jejunum.

Although there was an endovascular aneurysm repair (EVAR) in the medical history of another patient diagnosed with PAEF, the fistula was located between the left iliac artery as a result of the expansion of the existing sigmoid tumour. There was no EVAR stent graft in this region. In this patient, AEF did not originate from the graft, but from the growth of the tumour on the intestine towards the vascular structures. The fistula localisations were at the level of the duodenum in all patients with SAEF. The main symptoms of patients on initial admission were poor general condition, abdominal pain and GI bleeding. Melena was present in all patients. Fistulas in the upper GI tract (duodenum 6/8) were mostly hematemesis as a bleeding clinic, while fistulas in the lower GI tract (colon 2/8) were in the form of hematochezia. One patient had Behçet's disease, one patient had colon cancer, and one patient had gynaecologic malignancy as concomitant diseases. Two patients had renal insufficiency, and all patients generally had pulmonary, blood pressure and diabetic problems (Table I).

Microorganisms, such as *Staphylococcus hominis*, *Enterococcus faecalis*, *Enterococcus faecium*, *Escherichia coli*, *Candida albicans*, *Streptococcus anginosus*, *Enterobacter cloacae*, *Prevotella denticola*, and *Acinetobacter baumannii*, were identified from patients' tissues, surgical materials and catheters. In all but one patient, infected grafts were removed, extra-anatomical bypass procedures were performed, and bowel repairs, such as omental filling and fistulectomy with duodenojejunostomy in the area of the fistula, were performed. Only one patient underwent bowel repair with an open surgical method following an endovascular vascular repair in the form of a hybrid procedure. The 30-day hospital mortality rate for all patients was 50% (Table II).

Computed tomography (CT) angiography was used in the final diagnosis of the patients, and oesophago-gastroduodenoscopy and/or colonoscopy were performed in appropriate patients to confirm the diagnosis (Figures 1 and 2).

Discussion

AEF is a rare and highly fatal GI bleeding cause, even in the hands of experienced surgeons. AEFs, occurring as primary or secondary, most commonly develop after open or endovascular aortic procedures.⁸ If a connection between the natural aorta and part of the GI system is present, it is defined as a PAEF and if a connection

Table I: Clinical characteristics of patients with AEF

Sex/age	PAEF/SAEF	Prior aortic surgery	Fistula location	GI bleeding clinic	Comorbid diseases
Case 1	SAEF	+(Open surgery)	Duodenum	Melena + hematemesis	Behçet's disease + COPD + HT
Case 2	PAEF*	+(EVAR)	Colon	Melena + hematemesis/hematochezia	Colon Ca + leukaemia + COPD + HT
Case 3	SAEF	+(Open surgery)	Duodenum	Melena + hematemesis	HT + DM + renal failure
Case 4	SAEF	+(EVAR)	Duodenum	Melena + hematemesis	HT + DM + COPD
Case 5	SAEF	+(EVAR)	Duodenum	Melena + hematemesis	HT + COPD
Case 6	SAEF	+(Open surgery)	Duodenum	Melena + hematemesis	HT + DM + COPD + renal failure
Case 7	SAEF	+(Open surgery)	Colon	Melena + hematemesis/hematochezia	HT + COPD
Case 8	PAEF	None	Duodenojejunal junction	Melena + hematemesis	Gynecologic malignancy + COPD + HT

* The fistula was located between the native left iliac artery as a result of the expansion of the existing sigmoid tumour. Since there was no EVAR stent graft in this area, this patient was classified as PAEF. AEF – aortoenteric fistula, DM – diabetes mellitus, HT – hypertension, PAEF – primary aortoenteric fistula, SAEF – secondary aortoenteric fistula, COPD – chronic obstructive pulmonary disease, EVAR – endovascular aneurysm repair

Table II: Patients' surgical details, microbiological identification and clinical outcomes

Surgery	Microbiological identification	30-day hospital mortality
Case 1	Infected graft removal + axillobifemoral bypass + bowel repair	<i>Staphylococcus hominis</i> Discharged from hospital
Case 2	Iliac artery ligation + femorofemoral bypass + bowel repair	<i>Enterococcus faecium</i> + <i>Escherichia coli</i> + <i>Candida albicans</i> + Died
Case 3	Infected graft removal + axillobifemoral bypass + bowel repair	<i>Prevotella denticola</i> + Died
Case 4	Endovascular repair + bowel repair	<i>Acinetobacter baumannii</i> + Died
Case 5	Infected graft removal + axillobifemoral bypass + bowel repair	<i>Streptococcus anginosus</i> Discharged from hospital
Case 6	Infected graft removal + axillobifemoral bypass + bowel repair	<i>Morganella morganii</i> + Died
Case 7	Infected graft removal + axillobifemoral bypass + bowel repair	<i>Enterococcus faecalis</i> + <i>Enterococcus faecium</i> + <i>Escherichia coli</i> + <i>Enterobacter cloacae</i> + <i>Bacteroides fragilis</i> + <i>Candida albicans</i> Discharged from hospital
Case 8	Primary repair + femorofemoral bypass + bowel repair	<i>Enterobacter cloacae</i> + <i>Enterococcus faecium</i> Discharged from hospital

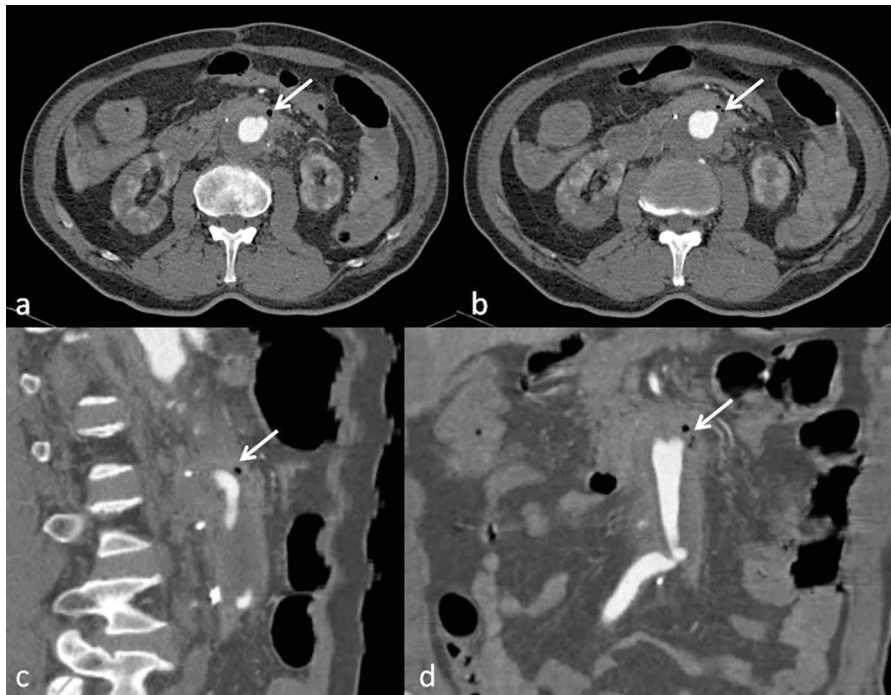


Figure 1: A patient (case 3) who had undergone aortobifemoral bypass 8 months ago at another outside centre presented with complaints of active GI bleeding and weakness in the left lower extremity. In arterial phase-contrast CT, axial (A, B), sagittal (C), and coronal (D) cross-sectional images. Aneurysm formation is seen at the infrarenal level in the abdominal aorta and surrounding thrombus material, as well as air bubbles in the left anterolateral thrombus material at the aortic-graft junction at this location (arrows). In addition, CT images show a tethered adjacent duodenum, soft tissue thickening and fluid around the aorta. The anterior section of the aorta appears to become uncertain, suggestive of an aortoenteric fistula.
GI – gastrointestinal, CT – computed tomography

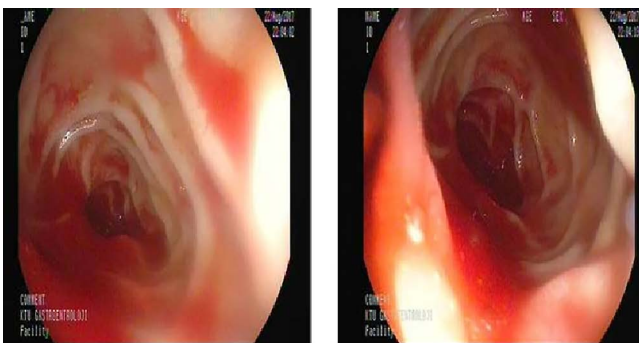


Figure 2: Oesophagogastroduodenoscopic images of a patient (case 6) complaining of GI bleeding. It has been described as an appearance suggestive of an aortoenteric fistula between the second and third parts of the duodenum.
GI – gastrointestinal

between an intestinal structure and a previous aortic graft is present, it is classified as a SAEF. If the aortic graft becomes infected, SAEF may develop in 20–45% of cases.⁹ The annual incidences of PAEF and SAEF in different series in the literature are approximately 0.04–0.07% and 0.36–2%, respectively.^{6,10,11} The SAEF rate after EVAR for atherosclerotic aneurysmal disease is less than 0.5% in the literature. This rate is lower than the AEF rate observed after open aortic repair (1.6%).^{12,13}

The experience gained in our cases and the review of the literature have provided us with important and instructive information. However, the ideal level of diagnosis, follow-up and treatment has not yet been achieved. One reason is that most of the articles in the literature are case reports and the case series include a relatively small number of patients

and do not include all possible complications of this clinical condition.

Because AEF is an unexpected cause of GI haemorrhage, medical history is critical to diagnosis. Early diagnosis and intervention have a beneficial and preventive effect on mortality and morbidity at this stage.¹⁴ Very few of the patients with AEF have massive bleeding. Patients with GI bleeding, regardless of signs of systemic infection, are usually in a poor clinical condition.¹¹ The most critical aspect of effective surgical treatment is diagnosis during the precursor haemorrhage before the fatal haemorrhage. Hypotension and hematemesis are the most common symptoms of this interval, which can last from a few hours to several months. When the diagnosis and intervention are delayed, problems may progress rapidly, like a snowball becoming an avalanche. As a result, patients and doctors can experience desperate moments. Unfortunately, this situation is analogous to a nightmare.

If the patient with suspected AEF is unstable and bleeding massively, diagnostic tests can be ominously futile. Urgent laparotomy is required to control bleeding and diagnose or rule out AEF in these patients. If patients are stable, they can be evaluated by endoscopy or CT for diagnosis. However, oesophagogastroduodenoscopy can detect AEF in only about half of these patients.¹⁴ Scintigraphy and 18F-fludeoxyglucose (FDG)-positron emission tomography (PET)/CT are nearly perfect for diagnosing AEF but are technically impractical. Magnetic resonance imaging (MRI) has not been the preferred method of diagnosis because the screening time is longer and more technical expertise is required, and pulse artifacts and perigraft gas cannot be distinguished from aortic wall calcification.¹⁵

In recent years, CT has become the most commonly used procedure to confirm the diagnosis. The presence of gas or fluid around the graft or aorta can be revealed on a CT scan with contrast enhancement, which can help the diagnosis. A decrease in the periaortic fat plane, bowel wall thickness changes near the aorta, ectopic gas, tethering of adjacent thickened intestinal loops toward the aortic graft and a pseudoaneurysm in the anastomotic region are other symptoms that verify the diagnosis of AEF with CT. Aortic contrast extravasation into the bowel lumen is exceedingly uncommon, but it is the most distinguishing characteristic of AEF.^{10,14,16,17}

The third part of the duodenum occupies anatomically a fixed position in the retroperitoneal region between the ligamentum Treitz, the aorta and the superior mesenteric artery. The proximity of this part of the duodenum to the aorta and to the proximal anastomotic region of the graft favours the development of AEF.⁶ Our series also reflects this situation. In our six cases, AEF formation was observed in this defined area.

One of the most important critical factors affecting survival in these individuals is immunity against the infections. In patients diagnosed with AEF, empiric broad-spectrum antibiotic treatment should be started as soon as microbiological culture samples are obtained. Further patient management and surgical preparation should be completed without waiting for culture results.¹⁶ Microbiological culture results of the cases reveal not only gram-negative or gram-positive organisms but also mixed-type microorganisms. About 20% of the culture results are polymicrobial.² Various microorganisms were also identified in our cases and in accordance with the results of the antibiogram, broad-spectrum antibiotics were given to the patients.

The main principles of AEF treatment may include extensive surgery to restore vascular circulation, closure of the fistula tract and complete removal of the infected prosthetic material.¹⁸ Considering that the potential for graft infection decreases as the graft is removed from infected surfaces, we ensured vascular continuity in our patients with an extra-anatomic bypass.¹⁹ Simultaneous bowel repair, complete excision of the infected graft, and axillobifemoral bypass procedures have been reported in the literature in patients with AEF, reducing early mortality and shortening the duration of surgery and trauma.²⁰

Because the patency of extra-anatomic bypass grafts is low, it is appropriate to perform vascular reconstructions in these patients with bypass techniques in the anatomic position. For this purpose, graft materials, such as antibiotic or silver-coated infection-resistant prosthetic grafts, cryopreserved grafts, and autologous vein grafts, have been developed.^{20,21} Each has its advantages and disadvantages. The major disadvantages of antimicrobial prosthetic grafts are the particularly low efficacy against methicillin-resistant microorganisms and the low graft patency rates.^{22,23} Cryopreserved arterial allografts may not be available in all centres because of their high cost. In addition, 10% of these patients have the possibility of spontaneous rupture.²⁴ Although autologous saphenous graft (neoaortoiliac surgical reconstruction) is the most effective graft against infection, it has problems such as prolongation of surgical time in graft preparation, bleeding problems associated with the graft and clinical complaints of the extremity.²⁴

In recent years, treatment of AEF has been associated with high morbidity and mortality, so more minimally invasive options have been tried. Endovascular repair is usually

successful in the short term and has theoretical advantages with less surgical exposure and trauma, shorter operative time and less need for transfusion. EVAR may be used more frequently in patients who are not candidates for open surgery due to anatomic incompatibility or poor general health conditions caused by sepsis and haemorrhagic shock. However, the results show that in patients who are candidates for open repair, EVAR can be used as a bridging measure until definitive repair, especially in the presence of a septic condition.^{7,13,18,25}

The insightful findings which we obtained from our cases and literature review can be summarised as follows: (i) When patients who have undergone aortoiliac surgery or intervention are admitted to the emergency department with GIS bleeding, these patients should be considered AEF until proven otherwise. (ii) Early surgery or intervention should be planned for patients with a strong suspicion of AEF. (iii) Patients' haemodynamic and clinical status should be monitored closely if their GI bleeding is stopped, because of the risk of herald bleeding. (IV) Patients with suspected or diagnosed AEF should be started on broad-spectrum anti-infective therapy as soon as possible after microbiological samples are collected.

Conclusion

In conclusion, AEFs are life-threatening conditions in which delays in diagnosis lead to catastrophic bleeding, morbidity and mortality. Outcomes depend on the early diagnosis, the medical status of the patient, the severity of infection and the anatomic location of the affected aorta. Graft excision and extra-anatomic bypass surgery or in situ graft replacement are the standard treatments for AEFs. Endovascular repair, on the other hand, has recently emerged as a viable therapeutic option. A multidisciplinary approach, appropriate treatment planning and close follow-up after treatment lead to positive outcomes.

Conflict of interest

The authors declare no conflict of interest.




Funding source

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Ethical approval

The medical ethical committee of Faculty of Medicine, Karadeniz Technical University approved the study protocol (24237895-567) and declared the study as not covered under WMO (Medical Research Involving Human Subjects Act) and therefore informed consent was not required (2021/171).

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