Case Report

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Digestive hemorrhage and fever as a result of a double secondary aortoenteric fistula following the repair of a juxtarenal abdominal aortic aneurysm and an infection of the aortobifemoral bypass graft: a case report

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Introduction: A double secondary aortoenteric fistula (AEF) occurs in a patient who has had significant aortic surgery and is characterized by a direct connection between the gastrointestinal (GI) tract and the aorta at two separate sites.
Importance: During aortoc reconstructive surgery, the patient may present with a variety of unusual complaints, including fever and GI bleeding. These symptoms are indicative of problems, including the development of an aortoentric fistula, particularly when there is a double secondary fistula.
Case presentation: The patient was admitted to the hospital due to hematemesis, melena, and high-grade fever after undergoing synthetic grafting aortobifemoral bypass (anatomical reconstruction) and partial resection of the juxtarenal abdominal aortic aneurysm. Pus discharge and a double aortoenteric fistula in unusual sites such as the second-third portion of the duodenum and caecum are visible in upper GI endoscopy and computed tomography angiography. The patient underwent a two-stage open surgery, the first stage involving aortic limb graft exclusion and extra anatomical reconstruction, and the second stage involving graft removal, fistula management, and bowel repair. Then the patient spent a few days in the surgical intensive care unit before being discharged.
Clinical discussion: Primary and secondary AEF are the two categories of AEF. In patients who underwent aortic reconstruction surgery, the frequency of secondary AEF ranges from 0.36 to 1.6%. Due to the 8:1 injury ratio in the secondery AEF, men suffer more injuries than women.There are two types of fistula depending on whether or not the suture line is involved. The first form is graft enteric erosion, which excludes the suture line, while the second type is entric graft fistula, where the suture line is included. Most common site fistula is third and fourth part of duodenum and least common site is fistula formation in large bowel.
Conclusions: An uncommon complication is double secondary AEF following aortic reconstruction surgery. Since one of the most significant presentations an AEF patient can present with is major GI bleeding and sepsis, A delay in seeking immediate medical treatment could result in the patient's death. It should be emphasized that one of the mechanisms for AEF formation and a frequent cause of sepsis in patients is recurrent aortic graft infection following aortic reconstruction surgery.

Keywords: abdominal aortic aneurysm, aortic graft infection, aortobifemoral bypass, case report, digestive haemorrhage, double secondary aortoenteric fistula, endovascular aortic reconstruction

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Sponsorships or competing interests that may be relevant to content are disclosed at the end of this article.

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HIGHLIGHTS

- A double secondary aortoenteric fistula is one of the rare complications of juxtarenal abdominal aortic aneurysm repair.
- Digestive haemorrhage and sepsis are the most common presentations of double secondary aortoenteric fistula.
- The mechanism responsible for the formation of the aortoenteric fistula and the attempt to reduce its occurrence.

Introduction

A digestive haemorrhage may be a symptom of the potentially fatal condition aortoenteric fistula (AEF)[1]. There are two types of AEFs: primary and secondary. Direct communication

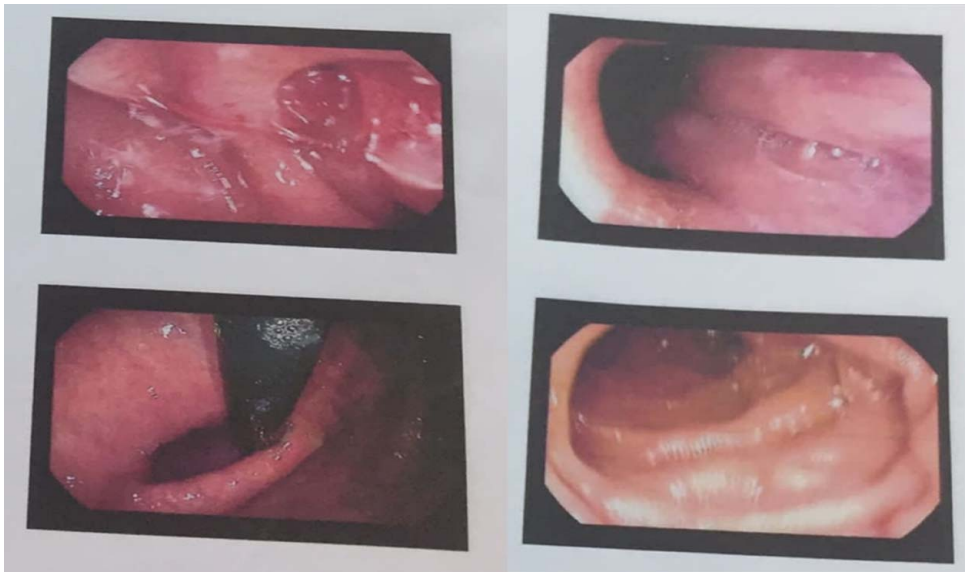

Figure 1. Upper GI endoscopy demonstrating fistula in the second-third portion of the duodenum in which pus discharge and blood were coming out. GI, gastrointestinal.

is possible via the primary AEF, an aorta-to-gastrointestinal (GI) tract link. An abdominal aortic aneurysm was once treated using a synthetic aortic graft. This condition, also known as secondary AEF, frequently results in deadly haemorrhages. The fatality rate without surgery is very nearly 100%[2–4]. Because double secondary AEF is so uncommon in practice, diagnosing it can be challenging. The most common methods for diagnosing AEF are computed tomography (CT) and EGD, with abdominal contrast-enhanced CT being the test of choice for the initial diagnosis of AEF[5]. The patient, in this case, had double secondary AEF (two fistulas originated from two different sites) following juxtarenal abdominal aortic aneurysm repair, which is a rare occurrence. Herein, we report an old male patient presenting to our emergency department complaining of GI haemorrhage and high-grade fever due to a double aortoenteric fistula. To the best of our knowledge, this is the first patient to have a double aortoenteric fistula in Palestine.

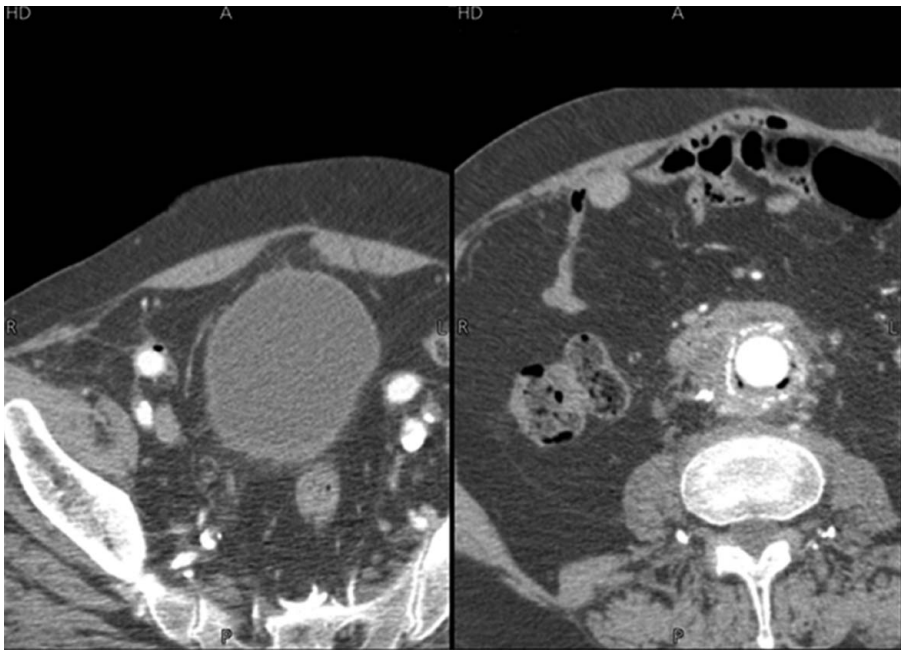

Figure 2. CT angiogram showing air in the wall of the aorta and air bubbles in caecum. CT, computed tomography.

Table 1

Summary of published 32 detailing case reports describing SAEF following AAA repair via open surgery and EVAR

Date	Authors and reference number	Age, sex	Clinical presentation	Initial treatment for aneurysm	Period[a]	Treatment for SAEF	Outcome	Pathological examination[b]	Estimated aetiology[c]
2021	Julia *et al.*[16]	68 M	Sepsis superimposed haemorrhagic shock developed after 1 l of lower gastrointestinal blood loss	EVAR	30 days	EVAR	Alive, Serial postoperative imaging to 18 months	–	Unclear
2019	Ryosuke *et al.*[17]	76 M	Abdominal discomfort	Open surgery	18 years	Aortic reconstruction, bowel repair	Alive, FU 12 months without development of any complications	+	Infection
2018	Arworn *et al.*[18]	42 M	Hematemesis, melena	EVAR	1 year 1 month	Aortic reconstruction, bowel repair	Alive at 9 month	+	Mechanical factor
2018	Alfawaz *et al.*[19]	62 M	Malaise, fever	Open surgery	7 years	Aortic reconstruction, bowel repair	Alive at 7 days	–	Unclear
2018	Jiang *et al.*[20]	85 M	Melena, tiredness, dizziness, fever	EVAR	2 years 4 months	Aortic reconstruction, bowel repair, debridement	Alive at 15 months	–	Unclear
2017	Hansen *et al.*[21]	75 F	Melena	Open surgery	2 years	Open surgery + EVAR	Alive at 12 years	–	Infection
2017	Dickfos *et al.*[22]	75 M	Abdominal pain, fevers	EVAR	1 year 2 months	Aortic reconstruction	Unclear	–	Infection
2017	Guevara-Noriega *et al.*[23]	65 M	Malaise	Open surgery	1 year 10 months	Aortic reconstruction, bowel repair	Alive at 2 years	–	Infection
2016	Kadhim *et al.*[24]	66 M	Confusion, fever	EVAR	15 years	Bowel repair, debridement	Alive at 12 months	+	Infection
2014	Zaki *et al.*[25]	75 M	Abdominal pain, hematemesis	EVAR	2 year 6 months	Aortic reconstruction, bowel repair	Died	–	Infection
2013	Jamal *et al.*[26]	60 F	Hematemesis, melena	Open surgery	6 years	Surgery	Died	–	Unclear
2011	Gullu *et al.*[27]	28 M		Open surgery	7 years	Suturing of aortic graft, bowel repair	Died	–	Unclear
2009	Grassia *et al.*[28]	72 M	Hematochezia	Open surgery	8 years	Aortic reconstruction	Alive at 2 weeks	–	Unclear
2009	McAloon *et al.*[29]	73 M	Lethargy, melena	Open surgery	10 years	None	Died	–	Unclear
2008	Hara *et al.*[30]	84 M	Hematochezia	Open surgery	14 years	Aortic reconstruction, bowel repair	Alive at 6 months	–	Recurrent aneurysm
2008	Geraci *et al.*[31]	59 M	Dyspepsia, vomiting	Open surgery	5 years	Bowel repair	Alive at 6 mo	–	Mechanical factor
2008	Bognar *et al.*[32]	67 M	Rectal bleeding	Open surgery	4 years	Aortic reconstruction, bowel repair	Alive at 24 days	+	Mechanical factor
2008	Alkim *et al.*[33]	35 M	upper gastrointestinal bleeding	Open surgery	2 year 3 months	1. Aortic reconstruction, bowel repair Alive, FU 15 months. 2. ABF, graft excision	Alive, FU 15 months.	–	Unclear
2008	Ogve *et al.*[34]	34 M	massive lower gastrointestinal tract bleeding	Open surgery	8 months	Suturing of aortic graft with omental wrapping, bowel repair	Alive	–	Unclear
2007	.Brountzos *et al.*[35]	85 M	Gastrointestinal bleeding	Open surgery	15 years	EVAR	Alive at 1 year	–	Unclear
2007	Tsunekawa *et al.*[36]	75 M	Fever, malaise	Open surgery	15 years	Aortic reconstruction, bowel repair	Alive at 1 month	+	Infection
2006	Heidemann *et al.*[37]	52 M	Hematochezia, hematemesis	Open surgery	6 months	Aortic reconstruction, bowel repair	Alive at 8 months	–	Unclear
2006	Maternini *et al.*[38]	73 M	Melena	Open surgery	15 years	EVAR	Alive	–	Unclear
2005	Mundal *et al.*[39]	82 F	Hematemesis	Open surgery	17 years	surgery	Died	+	Unclear
2004	French *et al.*[40]	68 F	Hematemesis	EVAR	1 year	Aortic reconstruction, bowel repair, debridement	Died	–	Infection

Table 1

(Continued)

Date	Authors and reference number	Age, sex	Clinical presentation	Initial treatment for aneurysm	Period[a]	Treatment for SAEF	Outcome	Pathological examination[b]	Estimated aetiology[c]
2002	Tomlinson *et a.f*[41]	90 M	Melena, abdominal pain	Open surgery	5 years	EVAR	Alive at 14 months	–	Unclear
2000	Makar *et al.*[42]	70 M	Epigastric discomfort	EVAR	4 months	Antibiotics	Died	–	Unclear
2000	Qzeren *et al.*[43]	35 M		Open surgery	5 years	Aortic reconstruction, bowel repair	Died		
1999	Karacagil *et al.*[44]	70 F	Melena, fever	Open surgery	14 years	Aortic reconstruction, bowel repair	Alive at 2 year	–	Unclear
1998	Yabu *et al.*[45]	77 M	Dyspnoea	Open surgery	10 years	None	None[d]	+	Mechanical factor
1993	Neergaard *et al.*[46]	69 M	Hematemesis, melena	Open surgery	8 years	Aortic reconstruction, bowel repair	Alive at 4 months	–	Unclear
1988	Kolke *et al.*[47]	45 M	left leg pain and tarry stool	Open surgery	21 months	Aortic reconstruction, bowel repair	Died	–	Unclear

AAA, abdominal aortic aneurysm; EVAR, endovascular aortic reconstruction; F, female; FU, follow-up; M, male; SAEF, secondary aortoenteric fistula.
[a]Period following AAA repairs.
[b]Whether or not a pathological examination was performed.
[c]Conceivable reason provided by the report's authors.
[d]Patient died immediately without receiving any care.

This case has been reported in line with SCARE criteria (see methods section).

Case report

A 63-year-old male married , smoker , non-drinker was admitted to the hospital for the evaluation of a fever and a small amount of melena and hematemesis. This was accompanied by generalized weakness, constant fatigue and shortness of breath. Note that the patient did not suffer from any dysphagia, dyspepsia, abdominal pain or any abnormal change of bowel habits. And the patient does not have any history of continuous use of NSAIDs, according to the patient, he used them when necessary. and he was found to have anaemia depending on a medical report done before hospital admission on the background of a medical history of the juxtarenal abdominal aortic aneurysm with synthetic grafting aortobifemoral bypass 1 year earlier, hypertension, type 2 diabetes mellitus, glaucoma, lens transplantation, and ischaemic heart disease with 6 times of catheterization with 4 stents. Note that the patient is committed to taking blood pressure, diabetes, lipid and anticoagulant medications. A medical workup, including an upper GI endoscopy (Fig. 1) and CT angiogram (Fig. 2), was done. There were a few millimetres of opening in the second-third portion of the duodenum, through which pus discharge and blood were coming out, which was compatible with an aortoenteric fistula. It also showed caecal air bubbles. On admission, the patient was transferred to the Surgical ICU as a case of sepsis (his vital signs were a blood pressure of 90/50 mmHg, tachycardia of 135 bpm, and a temperature of 38°C, and his lab report was white blood cell 5.56 with neutrophil 96.8, haemoglobin% 11.41, ESr35 mm/h, and C-reactive protein 185.4 mg/dl), managed with IV antibiotics, and referred to the surgical ward after patient stabilization. After a few days, the patient underwent the first stage of the operation, in which an exclusion aortic limb graft with endarterectomy was performed, then an axillofemoral bypass was done, and then a femoro-femoral bypass was done using polytetrafluoroethylene. Following surgery, the patient was transferred to the ICU for close monitoring. During the second stage of the operation, we found intestinal adhesion, an aorto-duodenal fistula at the second-third part, an aortocecal fistula with severe inflammation and unhealthy tissue, an infected aortobifemoral graft with pus discharge, and large infrarenal AAA (aneurysm remnants after juxtarenal abdominal aorta repair). Then we did infrarenal aortic neck control. That is, the remainder of the juxtarenal abdominal aortic aneurysm has been removed, along with the old infected aortobifemoral graft, closure of the infrarenal abdominal aorta with double layers of prolein, fistula management, primary repair of the second-third part of the duodenum, and right hemicolectomy with primary anastomosis. The patient was kept in the SICU post-operation for further intensive care management and daily labs, and the patient started on total parental nutrition, stabilized, and was transferred to the surgical ward, where he started on an oral diet gradually and tolerated it. A follow-up CT scan was done, which had acceptable results: no leak and intact vascular supply to the lower limb and bowel. then the patient was discharged, in good general condition, afebrile, and with stable vital signs.

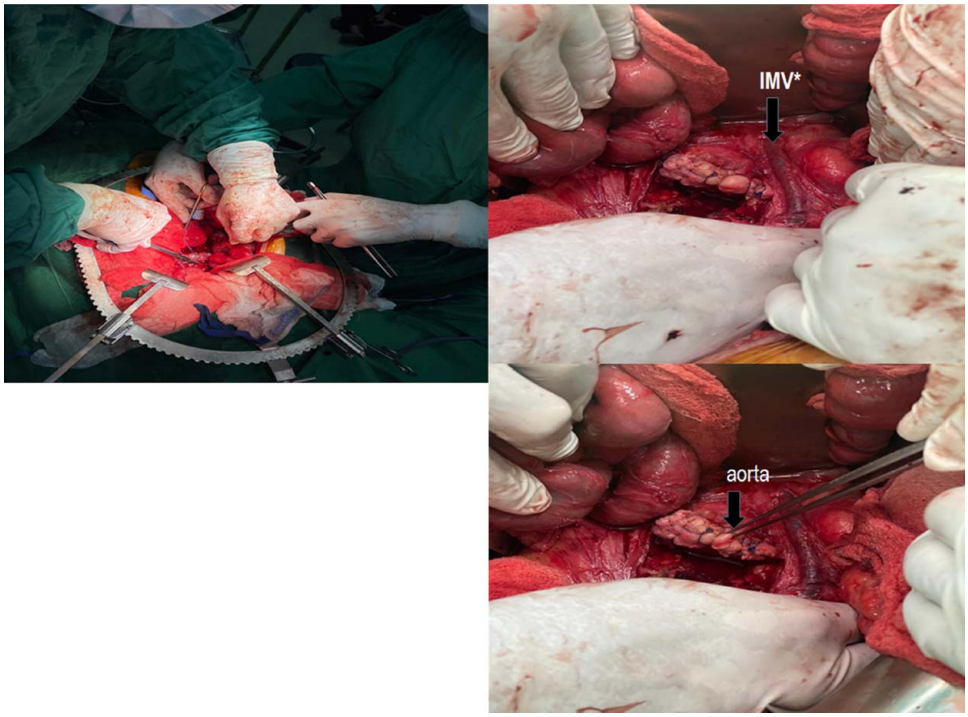

Figure 3. Intraoperative images taken after the remainder of the jauxternal AAA has had been removed, removal of old infected aortobifemoral graft, closure of infrarenal abdominal aorta with double layers of protein, fistula management, primary repair of the second- third part of the duodenum and right hemicolectomy with primay anastomosis. IMV, inferior mesenteric vein.

Discussion

AEF is divided into primary and secondary AEF. The incidence of secondary AEFs has surpassed that of primary AEFs[6]. According to reports, secondary AEFs happen in 0.36–1.6% of individuals who had their aortic condition surgically repaired as aortic aneurysm reconstruction[7–9]. Males are more likely to develop AEFs than females, and the average age of AEF development is 61 years. In general, males outnumber females in primary and secondary AEFs by a ratio of 3 to 1 and 8 to 1, respectively[10].

There are two different types of fistulas that emerge between the predominantly repaired or grafted aorta and the nearby bowel. The more typical type of secondary aortoenteric fistula (SAEF) is an enteric graft fistula, which is communication between the bowel and the damaged graft-aorta anastomotic site. The less common graft enteric erosion is caused by a breakdown of the enteric wall overlaying the graft, which leads to the graft being bathed in enteric contents and developing chronic graft infection without the involvement of a suture line. the uncommon SAEFs that can happen after procedures such as aortic endarterectomy, aortic repair for trauma, or aortic stump closure after removing an earlier aortic graft that takes place between the bowel lumen and an aortic suture line without the use of a prosthetic graft[7]. Our patient had an enteric graft fistula that resulted in communication between the bowel and the damaged graft-aorta anastomotic site. It is worth noting that the aneurysm that our patient was suffering from was 8 cm in diameter and unruptured when a juxtarenal AAA repair was performed, but when the aneurysm was removed, ~13 cm was left, meaning that not all of the aneurysm was removed. However, the repaired aorta remained free in the retroperitoneal area without being fixed to the peritoneum during surgery. This is one of the most important reasons for the recurrence of aortic graft infection and sepsis. One of the potential manifestations of aortic graft infection (AGI) is SAEF. According to reports, SAEF contributes 20–45% of AGI, and its incidence during the past three decades has been estimated at between 0.6 and 3%. Due to patients who were lost to follow-up, did not receive adequate follow-up, or were not informed about the late infection, the true incidence is surely greater[11–13]. Our patient had repeated hospital admissions for a case of sepsis and an aortic graft infection before he was diagnosed with double secondary AEF. The most frequent site of the AEF was the duodenum [62% of all cases and 77.6% of those reporting the precise location(s)], followed by the remaining small intestine, with the large bowel being impacted only rarely. According to the study, double (two co-existing) AEFs are incredibly uncommon[14]. With regard to the case we are presenting, the patient was admitted to the hospital on multiple occasions with sepsis and an AGI. Based on the tests and radiological images he had taken prior to being admitted, it was discovered that he had sepsis again and that he also had a fistula. Our patient also makes the distinction that he has double secondary AEF with the caecal region and the second-third portion of the duodenum. The fundamental process of fistula formation must be determined despite the significance of doing so. They are often diagnosed simply based on the clinical history or surgical findings; however, it has been proposed that either chronic infection of the graft, as in our case, or physical stimulation, such as from aortic pulsation pressure, contributes to their creation[15]. To our knowledge, only 32 case reports on SAEFs have been published (see Table 1). Among these, there is no double secondary AEF at rare sites, as it appeared in our patient, and in only

seven cases, the mechanism of fistula formation is infection. Therefore, the proportion of cases with double SAEF resulting from AGI is as rare as our case.

Conclusions

This case report describes GI bleeding due to a double SAEF at uncommon sites such as the caecum and second-third portion of the duodenum. Aortic graft infection (aortobifemoral bypass graft infection) was associated with its formation after juxtarenal AAA reconstruction due to communication between the aorta and the intestinal wall, indicating the importance of keeping a reconstructed aorta isolated from the intestine and giving prophylaxis antibiotics to reduce recurrence and the continuity of the follow-up to control and treat any complications that occur in the early stage. Clinicians should suspect AGI and double SAEF in patients with fever and melena after aneurysm surgery. Figure 3.

Methods

The work has been reported in line with the SCARE criteria.

Ethical approval

This study is exempt from ethical approval at our hospital.

Consent

Written informed consent was obtained from the patient for reporting this case. The consent is available for review on request.

Source of funding

The study did not receive any funding.

Author contribution

Writing the manuscript: A.A.A.J., M.A., O.N.D. Designing the figures: M.E., R.A., M.M. Reviewing and editing the manuscript: A.A.A.J., M.A. , O.N.D. ,M.E., R.A., M.M.

Conflicts of interest disclosure

There is no conflict of interest to declare.

Research registration unique identifying number (UIN)

NA.

Guarantor

Dr.Rajai Alhusseini.

Data availability statement

Not available.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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