

Aortoiliac Graft-enteric fistula presenting as gastrointestinal haemorrhage: A report on a complex-case management.

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Abstract

a 78-year-old patient presented with a life-threatening lower gastrointestinal bleeding secondary to an aortoiliac graft-enteric fistula into the sigmoid colon on the background of an adenocarcinoma and diverticular disease. Bridging endovascular stent followed by a second-stage graft explant and autologous vein reconstruction with a simultaneous anterior resection was successfully performed.

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Abstract

This case study discusses the staged management of a 78-year old patient presenting with life-threatening lower gastrointestinal (GI) bleeding secondary to an aortoiliac graft-enteric fistula (GEF) into the sigmoid colon on the background of an adenocarcinoma and diverticular disease. The patient had an aorto bi-iliac synthetic dacron graft repair of an abdominal aortic aneurysmal (AAA) some 20 years prior. Here, we present a case of successful endovascular treatment of massive haemorrhage, as a bridge to definitive second-stage dacron graft explant and autologous vein reconstruction with a simultaneous anterior resection.

Background

Arterio-enteric fistula (ArEF) is an umbrella term that encompasses various fistulations between the great arteries and the gastrointestinal (GI) tract, including aortoesophageal, aortogastric, aorto-enteric fistulas (AEF) and iliac artery-enteric fistula (IEF).

ArEF is a rare cause of potentially lethal GI bleeding and the vast majority appear to occur in the aorta, with few examples of IEF encountered in modern literature. These fistulas can occur via two separate pathological mechanisms. Primary ArEF are rare, occurring as a spontaneous communication between an artery and bowel from a combination of direct frictional forces and inflammatory processes [1]. The most encountered mechanism contributing to primary ArEF is aneurysmal formation, which is theorised to develop into a fistula from the repetitive mechanical forces exacerbated by cardiac pulsations and peristaltic movements [2]. This gradual degradation and erosion of the outermost layers of bowel and artery can also result spontaneously in other pathological circumstances; including tumours, diverticular disease, sepsis, syphilis and tuberculosis [3-6].

Secondary ArEFs are much more common given that they are an iatrogenic complication of open or endovascular AAA repair using synthetic graft. In this case, seeding of bacteria onto the synthetic graft is known to exacerbate the erosive mechanisms involved in fistula formation [2]. Hallet *et al* . have suggested a 1.6% chance of fistula formation after AAA graft repair [7]. The vast majority of ArEFs encountered in literature are between the abdominal aorta and duodenum due to their intimate anatomical association. IEFs are rarely encountered, but it appears that the majority occur secondary to pelvic surgery, malignancy, radiotherapy and infection [8].

In this case report we encountered a 78-year old patient, 20 years after an elective AAA Y-graft repair, presenting to emergency with haematochezia as a result of a fistula between the right common iliac artery (CIA) and sigmoid colon. The patient was known to have a sigmoid adenocarcinoma and diverticular disease from a colonoscopy three weeks prior to his emergency presentation. The rarity and complexity of this case necessitated a multidisciplinary staged approach to the management, both in the acute and elective setting, leading to a favourable outcome.

Case presentation

A 78-year old male, with a past medical history of atrial fibrillation (AF), psoriasis, hypertension and hyperlipidaemia, presented to the emergency department with syncope after passing 500ml of brisk fresh blood per rectum. 20 years prior, he had an uncomplicated elective infrarenal aorto bi-iliac graft repair of an AAA. His regular medications included bisoprolol, rivaroxaban and methotrexate at the time of admission.

The patient was hypotensive on presentation to the emergency department, with a blood pressure (BP) of 80/44 mmHg and heart rate of 124 beat/min.

Investigations

A colonoscopy three weeks prior to admission, due to weight loss and diarrhoea, showed a 4cm malignant-looking lesion in the sigmoid colon, which was tattooed, and several diverticula but no obvious fistulas were

seen (**Figure 1**). Histopathological analysis of the sigmoid lesion showed high-grade dysplasia.

An urgent triple-phase computed tomography angiogram (CTA) showed a ruptured pseudoaneurysm at the anastomotic junction of right aortoiliac graft limb and common iliac artery, which appeared to fistulate into the adjacent sigmoid colon. There was no obvious colonic metastatic disease (**Figure 2 A-C**).

Treatment

The patient was fluid resuscitated according to our hospital's massive transfusion protocol of 3000 units of Prothrombin Complex Concentrate (due to prolonged prothrombin time), 2 units of packed red cells, 1 unit of fresh frozen plasma, 1g tranexamic acid and 10mg of Vitamin K. Prophylactic intravenous Piperacillin-tazobactam was administered, given the likelihood of abdominal gut flora spreading onto the prosthetic material. His Rivaroxaban and Methotrexate were withheld.

Following initial stabilisation steps, and an urgent multidisciplinary team (MDT) meeting, it was decided that he would benefit from a temporising endovascular procedure to prevent further exsanguination, as a bridge to definitive surgical management.

Ultrasound-guided, percutaneous retrograde Right Common Femoral Artery access was secured, and an 8Fr sheath placed. The right internal iliac artery (IIA) was embolised with Concerto coils (Medtronic, Watford, UK). A 11x79mm balloon-mounted endoprosthesis (Viabahn VBX, Gore®, USA) was then deployed across the pseudoaneurysm, spanning from Iliac limb graft into the External Iliac Artery (**Figure 3**). After a period of in-hospital stabilisation, the patient was discharged home as per his wishes over the Christmas period, with a view to convalescence and work-up for definitive surgery. He was prescribed oral Co-Amoxiclav 625mg TDS as a suppressive regime given the likely infected prosthetic material. His Rivaroxaban and Methotrexate were withheld.

Colorectal MDT gave a predictive staging of T4b, N0, M0 sigmoid cancer, and the colorectal team advocated for an anterior resection with an end colostomy to mitigate any life-threatening risks that might arise from an anastomotic leak. The vascular surgery team proposed explantation of the aortoiliac Y graft and reconstruction using autologous deep vein. Alternative “fallback” options were discussed, including full or partial explantation with extra-anatomical bypass (femoro-femoral or axillo-femoral) or a more conservative approach with long-term suppressive antibiotics.

Satisfactory performance on Cardiopulmonary Exercise Testing (CPET) and Echocardiography gave objective support to the patient's cardiorespiratory fitness to proceed with complex major abdominal surgery. Venous duplex ultrasound examination confirmed sufficient superficial femoral vein (SFV) as potential autologous conduit for arterial reconstruction. It was agreed that both the colorectal and vascular components of surgery should happen simultaneously.

Soon after the New Year, the patient was reviewed in the vascular surgery and colorectal outpatient Clinics, where the proposed operative procedure, alternatives, risks and benefits were explained to the patient and his wife. He expressed his wish to proceed, and a date for surgery was fixed.

The patient underwent a joint procedure between the colorectal and vascular teams, via midline laparotomy. The finding of a fistula between right iliac limb anastomosis and the sigmoid colon was confirmed. The colonic tattoo appeared remote from the area of corruption. There was macroscopic impression of entire aortic graft involvement in an infective/inflammatory process. The procedure entailed: i) sigmoid colectomy with end colostomy, ii) right SFV harvesting and fashioning of Pantaloon bifurcated graft, iii) AAA Y-graft explantation, debridement of aortic sac, removal of Viabahn stent from CFA and arterial reconstruction with the autologous SFV graft, anastomosed to infrarenal aorta, the Left CIA origins, and Right EIA origin. iv) Omental coverage of the graft via a fenestration in the transverse mesocolon. Microbiological specimens were taken, including the prosthetic graft, pseudoaneurysm sac and associated thrombus and Viabahn stent graft.

There were no intraoperative complications during the procedure with an estimated blood loss of 2 litres over the 9-hour operation. The patient was transferred to the intensive care unit (ICU) and extubated after

24 hours. Microbiology results of the iliac stent graft demonstrated *Citrobacter koseri*, *E. coli*, *enterococcus faecium* and *bacteroides*. The aortic graft material showed evidence of *E. coli* and *enterococcus faecium*. Based on antibiotic susceptibility, the patient was commenced on teicoplanin, ciprofloxacin, and metronidazole, which were continued for 6-week course.

Outcome and follow-up

The patient stayed in the ICU for 4 days before being stepped down to the vascular ward for post-operative recovery totalling 30 days.

Postoperative complications included: i) ileus requiring total parenteral nutrition for a period of 21 days, ii) hospital acquired pneumonia, iii) wound dehiscence of the midline laparotomy 9 days post-operatively requiring an emergency relook laparotomy and primary closure, iv) right cerebellar embolic-type infarction 25 days post-operatively; patient continued on Rivaroxaban and did not require any further interventions given no focal neurological abnormalities and, v) seroma in patient's right thigh secondary to vein harvesting which was treated conservatively with compression stocking.

Histopathology results of the sigmoid colectomy diagnosed a moderately differentiated adenocarcinoma pT3, pN0, pMx, R0, with clear resection margins. Interestingly it also showed that diverticular disease was the most likely cause for the focal fistula, and was therefore completely unrelated to the tumour.

The patient went on to make a full recovery and was discharged home to complete the antibiotic regime. His methotrexate was eventually changed to an IL-23 inhibitor given the association between methotrexate and malignancy. On follow up 3- and 6- months, the patient has made a full recovery and remains clinically well. Surveillance colonoscopy, serum carcinoembryonic antigen (CEA) levels and CT scans were arranged as an outpatient according to NICE guidelines for follow up of colorectal cancer.

Discussion

This case report is the first case in literature describing an Iliac Artery Graft-Enteric fistula on the background of sigmoid cancer and diverticular disease. The acute endovascular 'bridging' procedure followed by planned definitive surgical intervention proved to be successful in the management of such complex and potentially life-threatening pathology.

Primary ArEFs are rarer than secondary ArEF, with an incidence of only 0.04-0.07%, compared to the 0.36-1.6% risk of developing a secondary ArEF after surgical treatment of aortic disease [7,10,11]. Encountering iliac artery-sigmoid fistulas in literature is rare, however the majority involve patients with a history of atherosclerotic aneurysm, pelvic malignancy or radiation [8,12]. An interesting feature of this particular case is the potential culprits that could have played a role in the development of a fistula; including diverticular disease, colorectal cancer and a difficult colonoscopy.

It is the authors' hypothesis that the patient developed tethering of a diverticular segment of sigmoid colon onto the graft- Right CIA. Colonoscopic investigation of the Red-Flag colonic cancer symptoms may have precipitated fistulation and pseudoaneurysmal degeneration, but this is difficult to be certain of, and may have happened in any case.

Histopathological analysis provided evidence of a diverticula being directly involved in fistula formation, and therefore was definitely a catalyst in the pathological process. Colorectal tumours are also associated with the development of fistulas [13], allowing us to assume that the adenocarcinoma may have influenced its formation via direct forces and inflammatory mechanisms. Colonoscopies don't have any direct correlating evidence in the pathophysiological development of ArEF, however khalaf *et al.* outlined a case report of a patient with an AEF into the sigmoid colon, which interestingly presented with lower GI bleeding during a colonoscopy

procedure [14]. In fact, colonoscopies have been associated with increased risk of rectocutaneous fistulas, which involves a similar pathophysiological process as ArEF [15]. The presentation of ArEF is associated with a triad of gastrointestinal bleeding, abdominal pain and a palpable abdominal mass - however studies have shown that this is seen in only 11% of cases [16].

CTA is the gold standard for initial assessment, with a reported sensitivity of 94% and specificity of 85% in recognising ArEFs. After initial resuscitation with fluids and oxygen, this patient was able to tolerate the investigation in order to aid diagnosis and formulate a strategy in management. The results of said investigation proved a high risk of death by exsanguination from the bleeding right CIA, and thus required immediate action to exclude the fistulous tract. Over the last few years, we have seen an emergence of endovascular techniques used for haemodynamic stabilisation, control of sepsis, bridge to definitive surgical repair and palliation for those at risk of major surgery. Danneel *et al.* have suggested using this ‘bridging’ procedure for all those with IEF who are considered high-risk for open repair [17]. We adopted this approach in the acute setting given that the CIA was a good target for endovascular stenting and that the patient would benefit from a period of stabilisation and planning for major complex definitive procedure to treat both the graft infection and sigmoid tumour. As a result, this time-saving approach has allowed for the patient to recover from the haemodynamic compromise, undergo preoperative investigations and have reduced risks from both local and systemic infection [17-19]. Given the assumption that the aorto bi-iliac graft was infected due to contact with the gut microbiota, the “gold-standard” of complete explantation and reconstruction with autologous venous conduit was considered the preferred approach [20, 21]. Numerous studies have found better outcomes when using autologous vein grafts rather than further synthetic graft material during reconstruction. Cryopreserved allografts, silver coated grafts, rifampicin bonded polyester grafts, or bovine pericardium are alternative options, but considered inferior to autologous deep vein in this case [22- 28].

Learning Points

- IEF is a rare cause of lower GI bleeding and can cause severe life-threatening consequences. A high degree of clinical suspicion is needed in patients with previous aortic surgery given the increased likelihood of secondary ArEFs.
- Using endovascular techniques in the acute setting as a staged approach can be extremely effective in maintaining haemodynamic stability and buying time for a safer elective procedure of total graft explantation and autologous vein reconstruction.
- Such complex cases are likely best managed in high-volume tertiary centres where multidisciplinary expertise is readily available.

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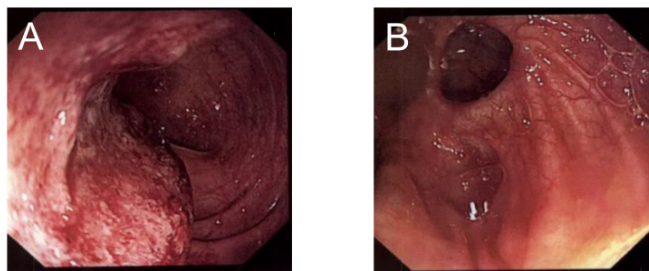


Figure 1: Colonoscopic findings. Colonoscopy showed: A) 4-cm malignant looking lesion in the sigmoid colon and B) evidence of diverticular disease extending into the hepatic flexure.

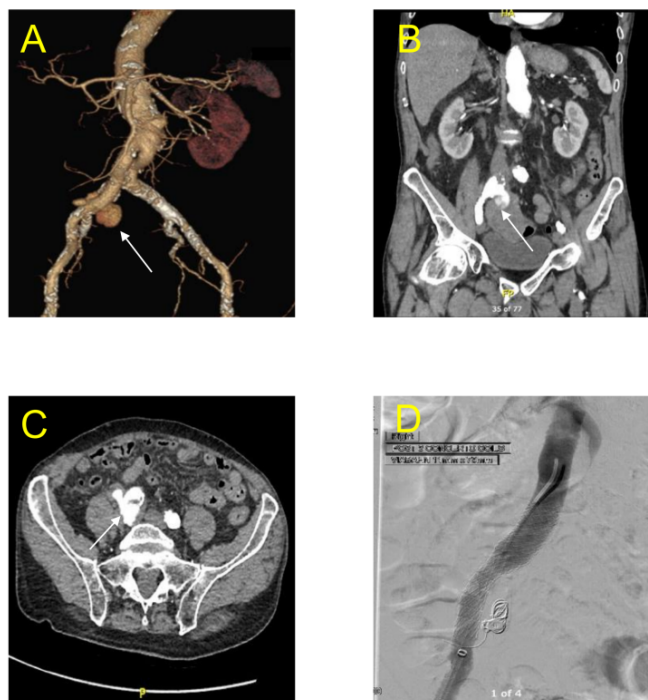


Figure 2: CT findings and stent insertion. A) Digital subtraction CT angiography image demonstrating right CIA anastomotic pseudoaneurysm (white arrow). B) Coronal CT image demonstrating the site of fistulation into the sigmoid colon. C) Axial image demonstrating the anatomy of the pseudoaneurysm. D) Image from angioplasty procedure demonstrating exclusion of the pseudoaneurysm using an endoprosthesis (stentgraft).

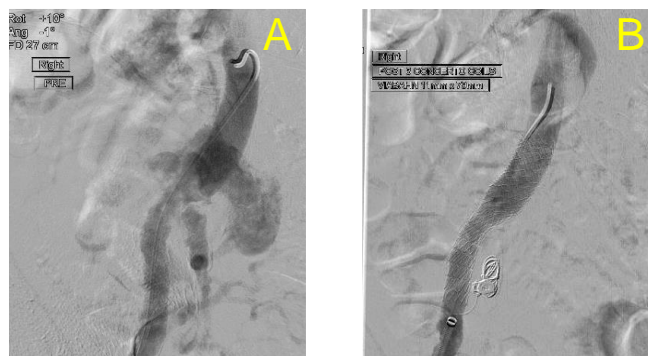


Figure 3: *Angioplasty* - Image from angioplasty procedure demonstrating : A) the pseudoaneurysm and B) the exclusion of the pseudoaneurysm using an endoprosthesis (stentgraft).