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## Case Report

## Secondary aortoenteric fistula—A fatal rare case involving the rectum ☆,☆☆

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## ABSTRACT

Aortoenteric fistula (AEF) is a rare but potentially fatal condition causing massive gastrointestinal bleeding. It is defined as fistulous communication between the gastrointestinal tract and the aorta which is sub classified into primary and secondary. Primary AEF refers to communication between a native aorta and the gastrointestinal tract. Secondary AEF is a communication between a reconstructed aorta, which includes open or endovascular repair, with the gastrointestinal tract. We herein describe an unusual case of secondary AEF in an 88-year-old gentleman. Our case is unusual as secondary AEF involving the rectum has rarely been reported in the literature.

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### Introduction

Aortoenteric fistula (AEF) is a rare and life threatening condition associated with massive gastrointestinal bleeding. AEF can be classified into primary and secondary with primary arising de novo in patients with no history of previous aortic surgery or trauma [1]. Secondary AEF are those which develop following any aortic reconstruction. Secondary AEF are more common in comparison to primary AEF but are never-

theless rare with an incidence of 0.6%-2% post abdominal aortic aneurysm (AAA) repair [2].

The duodenum is the most common site for fistulation accounting for 83% of reported cases of secondary AEF. Owing to the proximity of the infrarenal abdominal aorta, the third portion of the duodenum is the most commonly affected site. Other locations include the oesophagus, jejunum, appendix and sigmoid colon [3]. To the best of the authors knowledge, only 1 other case of secondary AEF involving the rectum has been published in the literature [4]. The involvement of the

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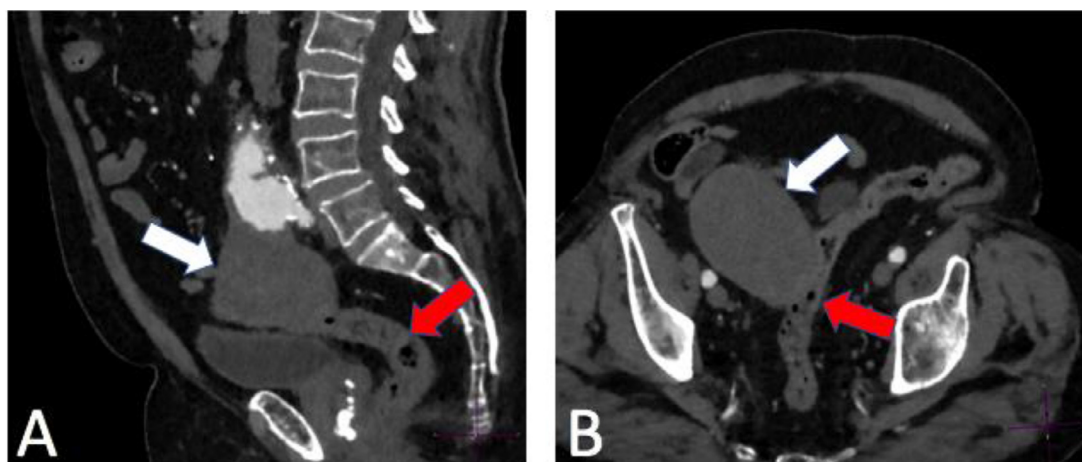
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**Fig. 1 – CT sagittal (A) and axial (B) reconstruction images performed 2 years prior to the described presentation showing a thrombosed pseudoaneurysm at the level of the aortic bifurcation (white arrow) and proximity to the rectum (red arrow).**

rectum is a rare entity due to the fact the rectum is the furthest enteric element from the abdominal aorta.

### Case report

An 88-year-old gentleman presented to the Accident and Emergency Department with rectal bleeding and altered bowel habit for an unspecified number of months. He described episodes of small volume bright red rectal bleeding with occasional episodes of blood mixed in with stool. The episodes of bleeding resolved spontaneously and he did not seek for medical attention initially. He also reported 2 stone (12.7 kg) weight loss in the same period with lethargy and increasing shortness of breath on exertion. He occasionally experienced crampy right lower quadrant abdominal pain.

His past medical history included a sigmoid colectomy for bowel adenocarcinoma (22 years prior to the described presentation) and a history of open AAA repair (28 years prior to the described presentation). Subsequent follow up CT imaging (Fig. 1 A and B), performed 2 years prior to the patient presenting to the Accident and Emergency department showed the formation of a large thrombosed pseudoaneurysm at the lower end of the aortic graft at the level of the aortic bifurcation measuring 10 cm x 6 cm in maximal diameter (AP x Transverse). On review of the CT imaging at that time, a multidisciplinary team including vascular surgery, deemed conservative management was appropriate given the patient's comorbidities and the limited number of safe vascular surgical options available to treat the pseudoaneurysm.

On examination, he appeared underweight with pallor. Observations were stable with BP 107/68, heart rate 99 bpm and temperature 35.8°C. Abdominal examination revealed a right iliac fossa mass. Fresh blood mixed in with stool was identified on rectal exam. No rectal masses were palpable.

Initial blood tests showed haemoglobin 96 g/L (baseline >100 g/L), eGFR 35 mls/min/1.73 m<sup>2</sup> (baseline >50 mls/min/1.73 m<sup>2</sup>) and mean corpuscular volume 76 fl. Co-

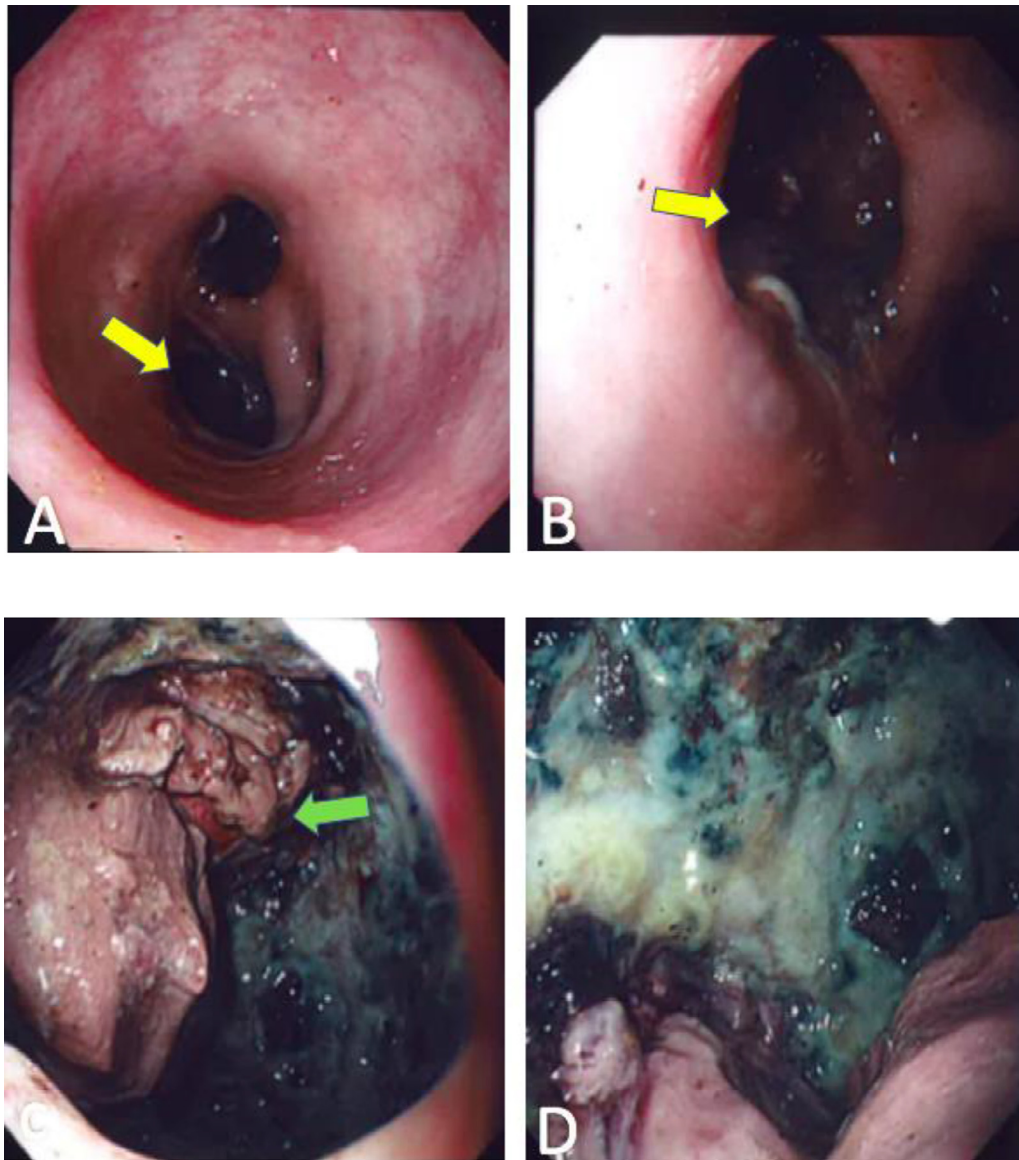
agulation screen was normal. White cell count was normal  $7.6 \times 10^9/l$  with raised c-reactive protein of 101 mg/L. No infective source was identified on urinalysis or chest x-ray.

Based on the patient's symptoms and baseline investigations, a working diagnosis of reoccurrence of bowel carcinoma was established. The patient was admitted into hospital under the care of the gastroenterology team. He was commenced on broad spectrum antibiotics and intravenous fluids. Following consultant post-take review, a CT scan of the abdomen and pelvis, gastroscopy and flexible sigmoidoscopy were requested.

Gastroscopy demonstrated changes consistent with atrophic gastritis within the body of the stomach. On sigmoidoscopy, a small blind ending lumen with a large mucosal defect was identified approximately 15 cm from the anal verge. The blind ending lumen contained a mixture of faeces and clotted blood products (Fig. 2 A-D). Appearances were deemed consistent with a longstanding bowel perforation.

Urgent CT abdomen and pelvis with oral and intravenous contrast in portal venous phase was performed following endoscopy. It showed an air and fluid level within the pseudoaneurysm wall with associated surrounding inflammatory changes. There was a loss of fat planes between the rectum and pseudoaneurysm with no appreciable IV contrast within the wall of the pseudoaneurysm or rectum. Appearances were felt to represent a fistula between the thrombosed aneurysm sac and upper rectum (Fig. 3 A-C).

After the diagnosis of AEF was confirmed on CT, palliative care measures were commenced as a safe surgical intervention was not available. On day 5 of admission, the patient became unresponsive and suffered a large volume bright red rectal bleed. Immediate resuscitative measures were initiated including intravenous fluids, blood transfusion and IV tranexamic acid. On review by the gastroenterology team, attempts of resuscitation in the event of cardiac arrest was deemed inappropriate. Comfort measures were initiated and the patient passed away approximately 4 hours after becoming unresponsive. Autopsy was not performed as the underlying cause of death was known.



**Fig. 2 – (A&B) Sigmoidoscopy images showing a small blind ending lumen (yellow arrow) at approximately 15cm from the anal verge. (C&D) Within the lumen, a large wall defect (green arrow) was noted with associated faeces and clotted blood.**

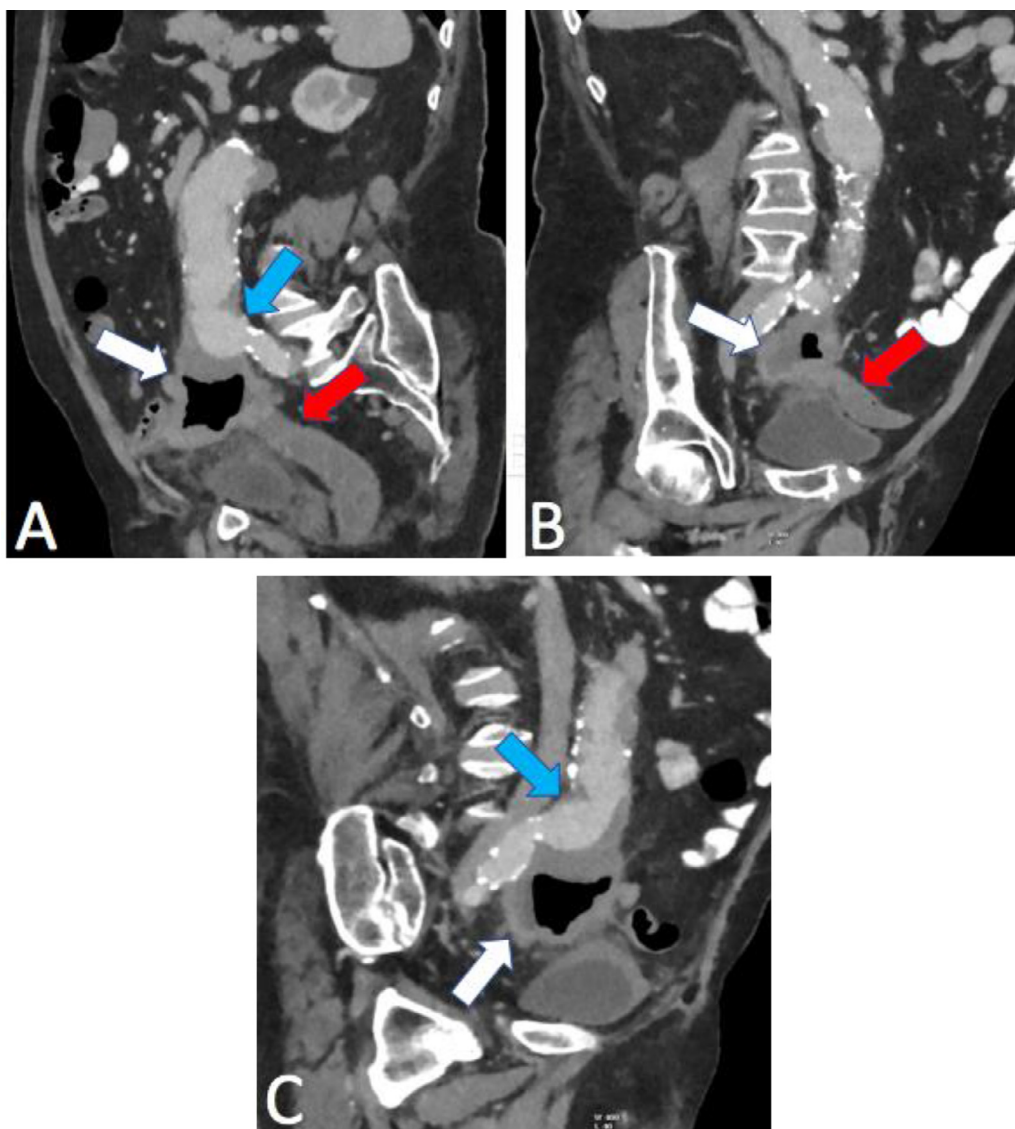
## Discussion

The exact pathogenesis of secondary AEF remains unknown but 2 mechanisms are commonly accepted. The first mechanism involves adhesion of an infected or inflamed graft to bowel wall. Graft infection leads to local inflammation, eventual breakdown and rupture into local structures. This theory is supported by detection of species of bacteria not usually identified in the intestine, such as *Staphylococcus*, in aortic prostheses in cases of secondary AEF. The second hypothesis postulates that the continuous pulsatile motion of the graft against the bowel wall leads to ischemia of the surrounding tissue and eventual erosion [5].

The clinical manifestations of AEF are often vague and nonspecific. The classical clinical triad of AEF was first de-

scribed by Sir Astley Cooper which consisted of abdominal pain, a pulsating abdominal mass and gastrointestinal bleeding [6]. Our patient reported vague abdominal pain and GI bleeding of unspecified duration. On examination, a mass was identified in the right iliac fossa. Our patient's symptoms correlate with the classical triad. However, this triad of symptoms remains a relatively uncommon clinical presentation only seen in 11% of cases [7].

The commonest reported presentation is GI bleeding which varies from self-limiting bleeding to overt hemorrhagic shock. The "herald bleed" is an episode of transient bleeding which is identified in 52%-75% of cases and often preclude a massive life threatening bleed [3,8]. Other reported clinical presentations include sepsis, abdominal pain, back pain or haemodynamic instability. Rarer presentations include lower limb ischemia, weight loss, anorexia and malaise. Indeed, our



**Fig. 3 – Curved multiplanar reconstruction CT images (A, B and C) showing an air and fluid filled pseudoaneurysm (white arrow) fistulating with the upper rectum (red arrow) at the level of the aortic bifurcation (blue arrow).**

patient presented with various nonspecific symptoms outside of the classic triad including weight loss and general malaise. An elevated CRP with no apparent cause was identified on the baseline investigations. The inflammatory markers improved on administering broad spectrum IV antibiotics.

The diagnosis of secondary AEF is challenging owing to variety of presentations, rarity of occurrence and variable interval between the initial aortic surgery and development of clinical symptoms related to AEF ranging from months to years [3]. Furthermore, despite the multitude of diagnostic tests available, no single investigation has high enough sensitivity or specificity in evaluation of secondary AEF. The diagnosis of secondary AEF is based on a strong clinical suspicion aided by readily available radiological investigations and GI endoscopy.

CT has wide ranging sensitivity (50%-94%) and specificity (85%-100%) [2,9]. The features of perigraft infection and secondary AEF overlap making differentiation between the 2

diagnoses challenging. Normal CT findings post graft insertion include perigraft soft tissue oedema/fluid, hematoma or ectopic gas. The perigraft soft tissue oedema or hematoma should resolve after 2-3 months of surgery however, any ectopic gas after 3-4 weeks should be considered abnormal and a strong suspicion for bowel fistulation should be considered [9]. Other shared features include loss of fat plane between bowel and the aorta, pseudoaneurysm formation and perigraft fluid. CT findings that correlate strongly with AEF include ectopic gas within the aorta (after 4 weeks), focal bowel wall thickening and breach of the aortic wall [2,9]. Extravasation of IV contrast material into the bowel lumen is the most specific feature of AEF but is extremely rare [9,10].

Multiphase CT angiography of the aorta is often the technique of choice. On unenhanced sequences, acute hematoma or partially thrombosed pseudoaneurysms are visualized as hyper attenuating fluid collections. CT arteriography is useful

for identifying disruption of the aortic wall or confirming the presence of AEF with extravasation of contrast material into the gastrointestinal lumen. Delayed imaging confirms endoluminal contrast leakage if AEF is present. Oral contrast is not routinely used however, the leakage of enteric contrast into the periaortic space is another rare but direct sign of AEF [11]. In our patient, CT findings included a gas and fluid level within the pseudoaneurysm and loss of fat plane between the aorta and rectum. Other imaging modalities including PET, MRI and Scintigraphy have been used as adjuncts with variable success in patients presenting with suspected AEF.

Upper GI endoscopy is recommended in patients with a history of AAA who present with stable upper GI bleeding [12]. The demonstration of alternative upper GI tract abnormalities does not conclusively exclude fistula formation and a high clinical suspicion should remain. Our patient presented with features of lower GI bleeding. As such, he underwent sigmoidoscopy which demonstrated a blind ending lumen with a large wall defect and associated clot products. The diagnostic yield for upper GI endoscopy is relatively modest at 40% [7], however, it is still a necessary investigation to rule out other commonly encountered pathologies causing upper GI patients.

## Conclusion

Secondary AEF is a rare complication of AAA repair and often patients present with vague symptoms of which GI bleeding is the most common presentation. No single investigation is reliably diagnostic and diagnosis of AEF relies on a high index of clinical suspicion in patients with known AAA or prior aortic intervention. CT imaging and endoscopy are the investigations of choice in patients who present with stable GI bleeding and a history of AAA repair. Specific CT findings that correlate with presence of AEF include ectopic gas within the aorta wall, local bowel wall thickening in the proximity of the aorta and breach of the aorta wall. Extravasation of IV contrast into

bowel lumen or extravasation of enteric contrast into the periaortic space are specific but rare findings in the diagnosis of AEF.

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