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Unusual site for primary arterio-enteric fistula resulting in massive upper gastrointestinal bleeding – A case report on presentation and management

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ABSTRACT

INTRODUCTION: Communications between an artery and the bowel are termed arterio-enteric fistulae. These are uncommon and mainly involve the aorta and duodenum. They can cause fatal haemorrhage. A primary aorto-enteric fistula has several aetiologies, one of which is post-radiotherapy.

CASE REPORT: 75-year old gentleman presented with acute upper gastrointestinal bleeding and haemorrhagic shock. He had a past history of right colonic cancer treated by resection and radiotherapy. At emergency gastroscopy he became critically unstable and the procedure was unsuccessful to achieve haemostasis. After resuscitation, a CT angiogram confirmed a right ilio-duodenal fistula between the right common iliac artery and duodenum. Interventional radiology was performed and a covered stent was inserted in the right common iliac artery. The patient recovered and was subsequently discharged from hospital. Three months later, he presented once again with similar massive haematemesis. Despite all efforts to stabilise him, he passed away a few hours after this second admission.

DISCUSSION: This case highlights what could possibly be a limitation of interventional radiology in providing definitive treatment for such a presentation. There are no set guidelines for the management of bleeding aorto-duodenal fistulae and literature is scarce. This makes it difficult to treat and the outcome is relatively unpredictable.

CONCLUSION: While minimally invasive radiological techniques are invaluable in many areas and life-saving in countless emergency bleeds, cases like these should ideally not be treated by stenting alone. It would be wise to follow arterio-enteric fistula bleeds by definitive open surgical repair.

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1. Introduction

An arterio-enteric fistula (AEF) consists of an abnormal communication between the aorta (or one of its branches) to any part of the small or large bowel [1]. It is a rare life-threatening cause for massive upper gastrointestinal bleeding and is fatal unless treated promptly. It involves the duodenum in more than 75% of cases, mostly the third or fourth duodenal segments [2].

The causes of AEF are classified as primary, secondary and the very rare idiopathic form. Primary aorto-duodenal fistula (PADF) was first described in 1822 by Sir Astley Cooper. It is a spontaneously forming fistula which may follow erosion by atherosclerotic aortic aneurysm (73%), invasion from malignancy,

peptic ulceration, trauma, foreign bodies, biliary stones, radiotherapy, inflammation and infection (mycotic) including syphilis, *Salmonella*, *Klebsiella*, spirochaetes, tuberculosis and fungi. The infra-renal aorta is affected in more than 75% of cases [3–5]. Secondary causes are often late complications of abdominal aortic aneurysm (AAA) repair including open and endovascular techniques [5]. Autopsy studies revealed an incidence of PADF of 0.04–0.07% [6].

PADF with upper gastrointestinal bleeding following radiotherapy was first reported in 1983 in the context of para-aortic and pelvic irradiation [7]. It is considered to be a rare cause of PADF, being more prevalent after external beam irradiation and having a latency period between 2 weeks and 25 years. The pathogenesis is unclear but radiation-induced chronic duodenal/bowel inflammation together with adjacent arterial pulsatile stress followed by erosion and fistulation into surrounding tissues is a plausible theory. The effects are amplified in the presence of duodenal ulceration and radiation-induced necrosis of the tunica media and adventitia of a nearby vessel [5]. A literature search was performed using

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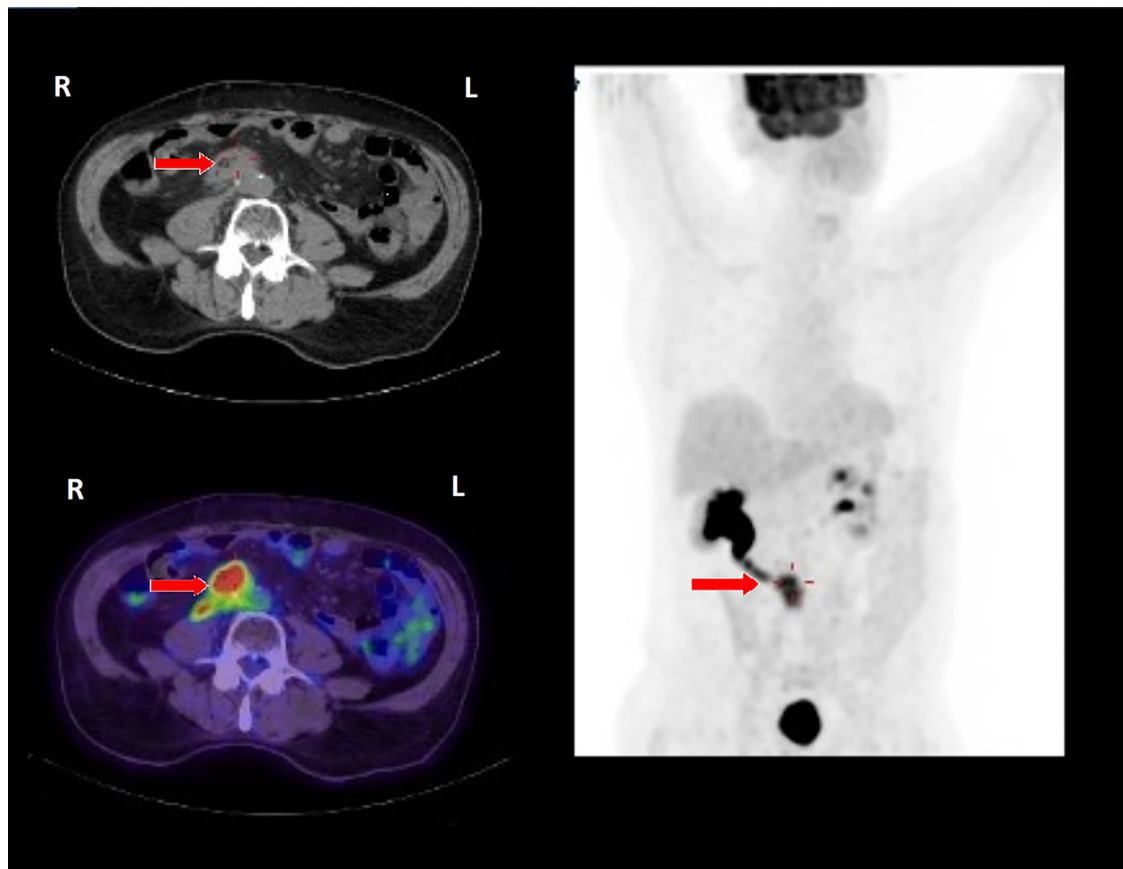


Fig. 1. Selected PET-CT images showing the metastatic right sided para-aortic lymphadenopathy that was targeted by external beam irradiation. This is involving the right ureter with resulting right sided hydronephrosis, as seen in the coronal PET image.

PUBMED and EMBASE databases. Following our literature review, no documented cases of post-radiotherapy fistulation between a common iliac artery and the duodenum were identified. This case report has been compiled in line with the SCARE criteria [8].

2. Case report

A 75 year old gentleman was brought by ambulance to the Emergency Department with haematemesis in July 2017. He was resuscitated with intravenous (IV) fluids. On examination, the patient was alert, but pale. His blood pressure recorded non-invasively was 100/60 mmHg, and his pulse was 80 beats per minute with a capillary refill time of less than 2 seconds. His oxygen saturation was 99% on room air. On physical examination, the patient's abdomen was soft with tenderness in the epigastric region. No rebound and no guarding was present when the abdomen was palpated. Urgent laboratory investigations showed a haemoglobin of 8.6 g/dL with a mean cell volume of 83.3 fL. His serum creatinine of 127 $\mu\text{mol/L}$ and urea was 10.9 mmol/L with eGFR of 48 mL/min/1.73m². His coagulation was normal (International Normalised Ratio: 1.06) and blood gases showed a pH of 7.44 and a lactate of 0.6 mmol/L.

His past medical history included hypertension for which he had been prescribed enalapril and amlodipine. He had a right hemicolectomy in 2011 for an ascending colon tumour (grade II, pT4N2Mx) followed by adjuvant chemoradiotherapy. A positron emission tomography (PET) scan which was done three months prior to this presentation had showed an increased tracer uptake in the interaortocaval lymph nodes at L3 and L4 level. Further enhanced uptake was confirmed by another PET Scan two months

prior to admission (Fig. 1). Due to this result, he underwent radiotherapy to these lymph nodes two months prior to this episode of haematemesis. The radiotherapy dose was 30 Gy given over 20 sessions.

Following resuscitation, the patient was transferred to the emergency theatre for an emergency oesophago-gastro-duodenoscopy (OGD). During rapid sequence induction, the patient had a hypotensive episode where the blood pressure dropped to 60/40 mmHg. During the OGD, a large clot was noted in the antrum and body of the stomach. Following washout of this clot, the pylorus was identified and the duodenum was intubated with the gastroscope. A large clot was found in the second part (D2) of the duodenum. Although washout of this clot was attempted, fresh blood continued to pool in the D2 (Fig. 2).

Since the patient remained hypotensive and definitive endoscopic therapy was not possible, a decision was taken to attempt angioembolisation. The initial angiogram showed bleeding through a fistula between the right common iliac artery and second to third portion of the duodenum (Fig. 3). The computed tomography (CT) scan also showed enlarged lymph nodes which were encasing the right common iliac artery and extending to lower aorta. A 10 mm wide and 5.8 cm long covered stent was deployed in the right common iliac artery by a consultant interventional radiologist (Fig. 4). Control angiography showed no evidence of residual haemorrhage (Fig. 5).

The patient was subsequently transferred to the Intensive Therapy Unit (ITU) where further resuscitation was performed. The patient was transfused red blood cell concentrates, platelets and tranexamic acid as per our local major haemorrhage protocol. Antibiotic regimen consisted of IV metronidazole 500 mg three

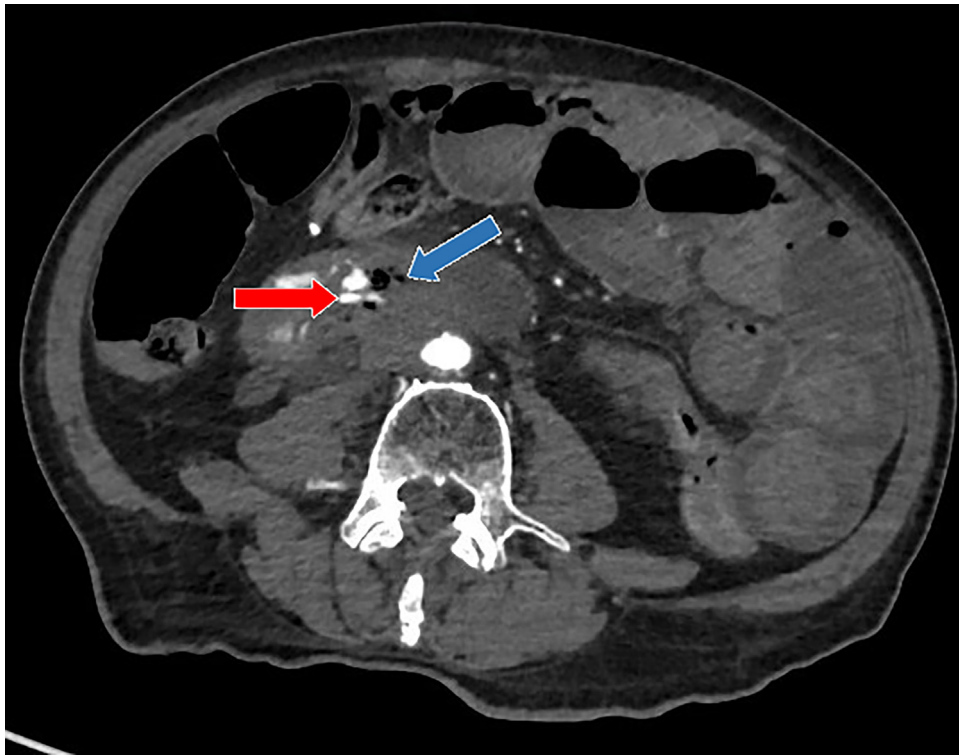


Fig. 2. Selected arterial phase axial CT image showing large volume active extravasation of contrast in a distended duodenum (red arrow). Blood products are seen in the visible loops of small bowel. A collection with bubbles of gas is seen between the duodenum and aorta (blue arrow).

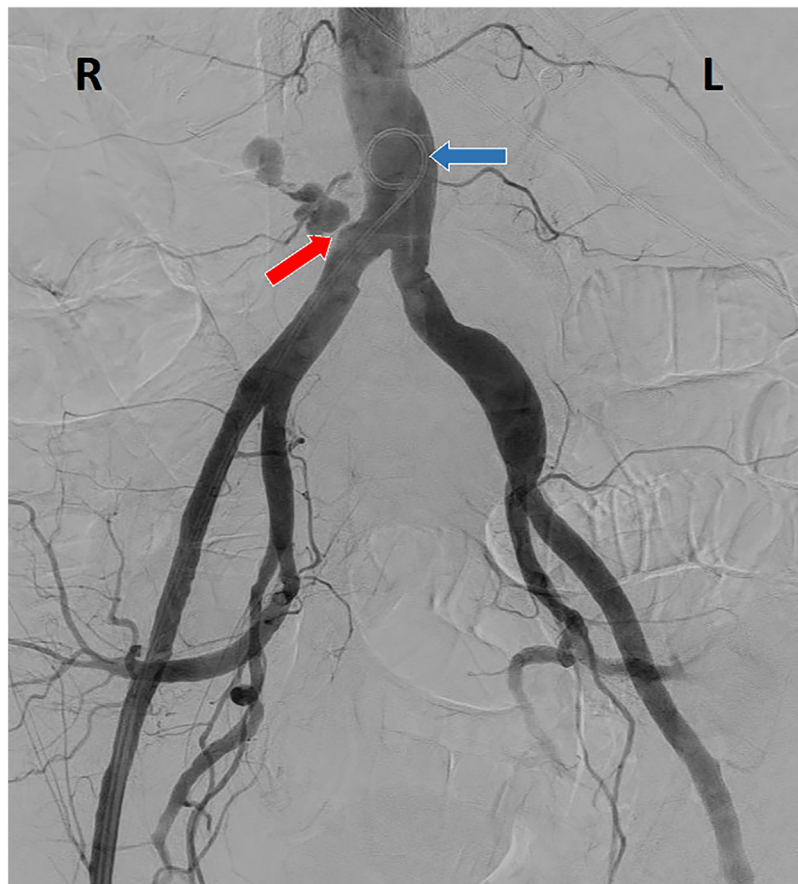


Fig. 3. Digital subtraction angiography showing a fistula between the medial aspect of the second/third portion of the duodenum and the superior portion of the right common iliac artery just below the level of the aortic bifurcation (red arrow). The pigtail tip of angiographic catheter can be seen just above the aortic bifurcation (blue arrow).



Fig. 4. Application of a 10 mm wide 5.8 cm long covered stent-graft.

times daily and ceftriaxone 2 g daily. He was extubated 3 days later and transferred to the ward. A haemoglobin drop to 8.5 g/dL was noted on day 5 post stent insertion which was managed conservatively. He continued to improve clinically and was discharged on day 14 post-admission at which point antibiotics were stopped.

A plan for open operative repair was made but after discussion with the patient and relatives, a conservative approach was opted for. The patient presented again 3 months later with re-bleeding from the fistula. He was unfit for emergency surgery and several attempts at minimally invasive re-stenting were unsuccessful. He succumbed to the event few hours after admission.

3. Discussion

The rarity of primary AEF limits individual and institutional experience to manage such a presentation [9]. As with most surgical emergencies, prompt diagnosis is key in determining outcome. Around a third of patients will die in the first 6–12 hours of symptom onset. Katsamouris et al in their 1994 case report mentioned a triad of symptoms: abdominal/back pain, gastrointestinal haemorrhage (maleana, haematemesis) and pulsatile abdominal mass.

Despite being considered the classical triad for AEF, less than 40% of patients present as such. Furthermore the triad is attributed to fistulae involving abdominal aortic aneurysms [4]. Less than 5% will present with haemorrhagic shock without previous episodes of minor bleeds [10]. The term 'herald bleed' is a sign where the patient complains of intermittent haematemesis or haematochezia up to 3 weeks prior to the massive haemorrhagic episode [11]. This sinister sign should raise suspicion of AEF even in the absence of positive endoscopic findings as it can be the only opportunity for early therapeutic attempts [12].

The management options in our case were limited by the patient's unstable condition, high surgical risk and uncertain diagnosis. Following the oesophagogastroduodenoscopy (OGD), interventional radiology was deemed the appropriate minimally invasive therapeutic option. As the main iliac artery was involved, a covered stent was the best way to establish vessel continuity and arrest the bleeding as opposed to embolisation.

The abdomino-pelvic CT included a pre-contrast phase followed by angiographic arterial and venous phases. CT with IV contrast is considered the best tool for diagnosing PADF in the stable patient (sensitivity 94%, specificity 85%) [13]. Oral contrast media should be avoided when imaging gastrointestinal bleeding as their diagnostic yield is low [4]. CT findings suggestive of PADF include retroperitoneal air or thrombus, loss of fat plane between aorta

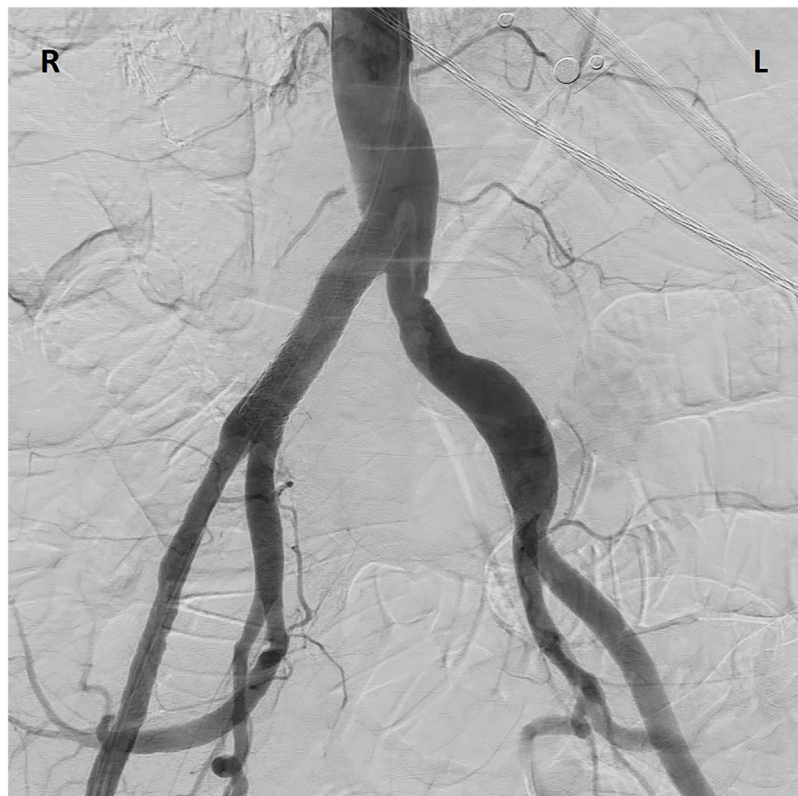


Fig. 5. Fistula was successfully closed while preserving patency of the aorta and right common, internal, and external iliac arteries.

and duodenum, and intestinal wall oedema. Extravasation of IV contrast into bowel further confirms the pathology. A negative endoscopy does not exclude PADF and many cases are missed with OGD alone. Ultrasound, magnetic resonance imaging and tagged white cell scans are seldom useful. Angiography requires a flow of more than 0.5 mL/min and only detects 30% of actively bleeding fistulae [14,15].

Richards et al., mention the benefit of permissive hypotension during the resuscitation of patients with bleeding aorto-duodenal fistula [11]. Surgical approaches changed over the years. Extra-anatomic bypass was the preferred option in the past [4]. Nowadays this is only recommended for cases with gross contamination or abscess around the fistula [3,5]. A more recent approach for radiation-induced PADF involves excision and reconstruction of the damaged duodenal wall, omental cover over the arterial repair and insertion of a prosthetic sleeve between bowel and vessel [5].

Specifically for PADF post-radiotherapy, primary repair without prosthetic grafting may be possible together with debridement of the inflamed and scarred tissue. Omental pedicled graft or protective sleeve placed between duodenum and the repaired vessel could also be considered as adjuncts to the procedure. Intra-operative haemorrhage is controlled by clamping the aorta just below the diaphragmatic hiatus [1,4,5]. Cultures from the arterial wall are taken and if positive, the patient should be given 4–6 weeks of antibiotics post-operatively [10,15]. Negative cultures require a one-week antibiotic regime. If primary repair of the small bowel is not possible, then resection and anastomosis is recommended. Primary suturing of small defects is appropriate, but omental flap interposition or duodenal switch are recommended for larger defects as there is a high risk of recurrence with suturing alone [11,14].

Endovascular aortic repair (EVAR) is a rapid, minimally invasive technique that can be employed in the unstable patient. Duvnjak et al., used coil embolisation to successfully treat a bleeding fistula between the left internal iliac artery and sigmoid colon in a patient who received radiotherapy for prostate and caecal cancer. There were no adverse events during the relatively short 5-month follow-up [13]. Leonhardt et al studied the outcome over a three-year period of five patients with abdominal AEF (four primary, one secondary) who underwent balloon occlusion and endovascular repair. The immediate success rate at stopping bleeding was 80% using stent grafts. Despite the initial success, 80% rebled after 2 weeks or longer. 30-day mortality was 40%, which doubled at 6 months. This supports the conclusion that EVAR should be considered only as bridging measure prior to definitive surgical repair [14]. An exception could be in the palliative setting where endovascular repair can be the only definite procedure [1]. As the risk of infection is high, patients require antibiotic cover following EVAR [14].

4. Conclusion

In cases of massive gastrointestinal haemorrhage, it is important to look for a history of abdominal/pelvic radiotherapy. A high index of suspicion is required to diagnose PADF as this may be missed with endoscopy alone. CT with IV contrast is considered the most effective method to diagnose PADF provided the patient is relatively haemodynamically stable [5,11]. Although surgery is the definitive treatment of PADF, in the immediate setting it can also be treated using a covered stent-graft. Such an approach might be advantageous given that significantly less time is needed to puncture the femoral artery and deploy the stent under local anaesthesia (total procedure time in our case was less than 10 minutes).

Conflict of interest

The authors declare no conflicts of interest.

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Ethical approval

The University of Malta Faculty of Research Ethics Committee declared that there is no need for ethical approval for this case report as there are no patient identifiers in the images or text.

Consent

Written informed consent was obtained from the patient's next of kin (wife) for the publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Anthony P. Dimech collected the data, was responsible for obtaining informed consent and ethical approval, did literature review and drafted the report. Matthew Sammut provided assistance in compiling and editing the report. He also helped in references. Kelvin Cortis provided images and details on the radiological procedure carried out on the patient. Nebosja Petrovic was involved in the initial assessment of the patient and proof-read the case report.

Registration of research studies

This case report was registered at: www.researchregistry.com Identifier: researchregistry3777.

Guarantor

Anthony P. Dimech.

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