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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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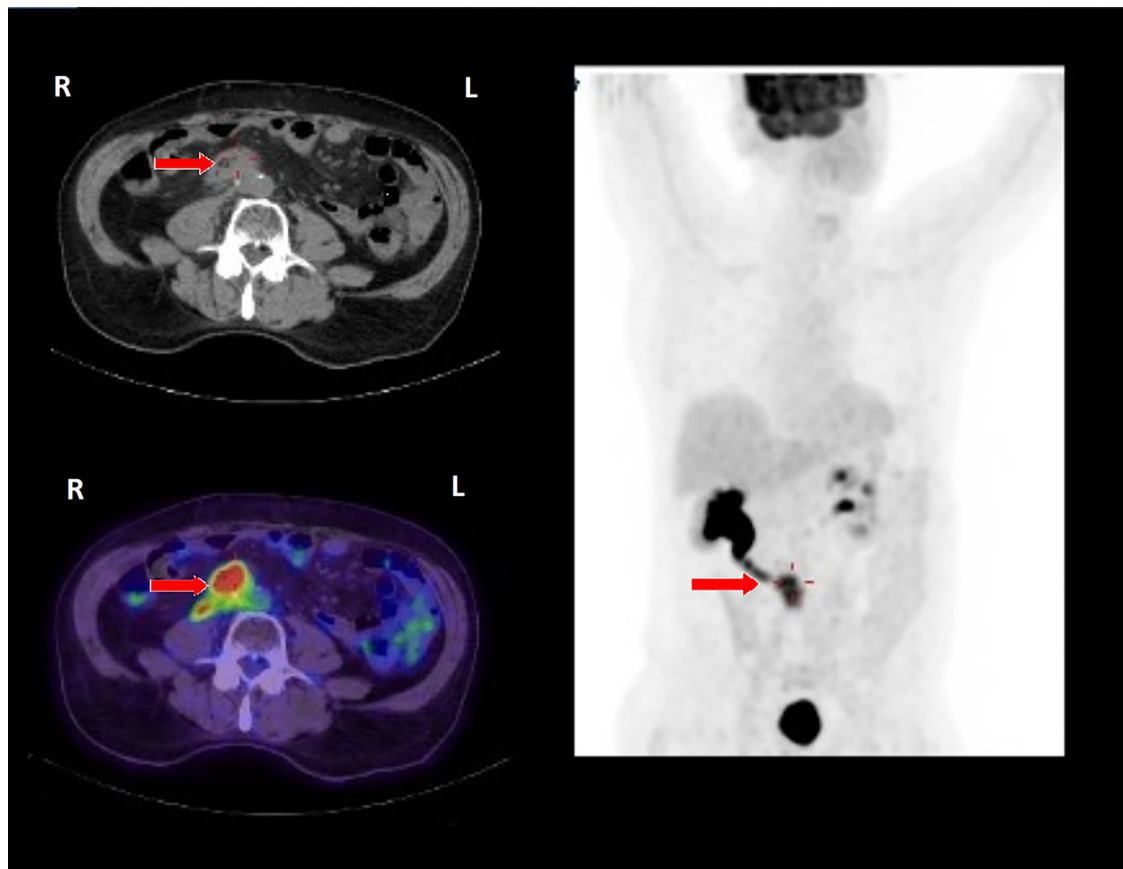


Fig. 1. Selected PET-CT images showing the metastatic right sided para-aortic lymphadenopathy that was targeted by external beam irradiation. This is involving the right ureter with resulting right sided hydronephrosis, as seen in the coronal PET image.

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 μ 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

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