



Vascular Interventions Case Report

Endovascular treatment of massive hemorrhage from a radiation-induced iliac artery-enteric fistula: A case report and review of the literature

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ABSTRACT

Arterioenteric fistula is a rare cause of massive gastrointestinal tract hemorrhage. It is defined as an abnormal communication between an artery and a bowel loop. Radiation-induced iliac artery-enteric fistula (IEF) is an extremely rare subtype with only a few cases reported in the literature. Here, we present a case of successful endovascular treatment of massive hemorrhage due to radiation-induced IEF.

Keywords: Aortoenteric fistula, Arterioenteric fistula, Iliac artery-enteric fistula, Interventional radiology

INTRODUCTION

An abnormal fistulous communication between the arterial tree and the gastrointestinal (GI) tract is a rare yet serious cause of GI hemorrhage. It is associated with a high mortality rate that approaches 100 % without immediate diagnosis and intervention.^[1]

Various terms have been used to describe this entity depending on the site of the abnormal connection, for example, aortoesophageal, aortogastric, aortoduodenal, aortoenteric fistula (AEF), and iliac artery-enteric fistulas (IEFs). Arterioenteric fistula (ArEF) is a more comprehensive term defined as a communication between a major artery and a bowel loop.

The majority of ArEFs involve the abdominal aorta. However, IEFs are rarely reported in the literature, with pelvic radiotherapy being a contributing factor. Here, we present a case of radiation-induced ArEF with a brief literature review of similar previously reported cases.

CASE REPORT

A 68-year-old woman of Chinese ethnicity presented to the emergency department with acute massive melena, hematochezia, hematemesis, and sharp colicky abdominal pain. She had a history of carcinoma of the cervix treated with radical hysterectomy and chemoradiotherapy. In addition, she was known to have ischemic heart disease.

On admission, she was hypotensive with a blood pressure of 72/42 mmHg and tachycardic with a heart rate of 112 beats/min. The patient was intubated and fluid resuscitated according to our hospital's massive transfusion protocol of four units of packed cells, one

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unit of pooled platelets, and one unit of fresh frozen plasma.

Urgent computed tomographic (CT) abdominal angiography revealed large amounts of intraluminal hyperdense blood products distributed within the small bowel loops [Figure 1]. In addition, there was active arterial contrast extravasation from the distal external iliac artery (EIA) into the distal ileal loops in the right lower abdomen through a fistulous tract [Figures 1 and 2].

The patient was immediately transferred to the interventional radiology suite for urgent catheter angiography. Pelvic angiogram revealed active contrast extravasation from the medial wall of the right EIA into the small bowel [Figure 3 and Video 1] confirming the suspected fistulous communication seen on the CT angiogram. Balloon tamponade of the right EIA was rapidly performed with a 5.5 Fr Fogarty balloon catheter (Edwards, Irvine CA, USA) to provide immediate hemodynamic control. Subsequently, a 7 mm × 5 cm Viabahn covered stent (Gore, USA) was deployed in the distal EIA successfully to exclude the fistulous tract. The size of the native external right iliac artery was 7 mm on the pre-procedure CT angiogram. A stent with a length of 5 cm was used to avoid crossing the right hip joint and make probable future surgical intervention easier. The stent did not require oversizing but to ensure a satisfactory sealing, it was post-dilated with a 7 mm angioplasty balloon. Post-procedural angiogram showed successful exclusion of the fistulous tract with no further angiographic evidence of hemorrhage [Figure 4 and Video 2].

The patient was subsequently transferred to the intensive care unit for close monitoring. She was commenced on intravenous antibiotic therapy. Post-procedure recovery was uneventful and she was discharged from the hospital several days later. There was no evidence of recurring fistulation, leak, or bleeding on her follow-up CT scans [Figure 5] up to 6 months following the procedure.

DISCUSSION

ArEF is a broad term that encompasses AEF, IEF fistula, and other subcategories. ArEF can be classified into primary and secondary types.^[1] The rarer primary ArEF was first described by Sir Astley Cooper in 1829. It occurs in patients who have never undergone aortic reconstruction/repair. The causes of primary ArEF include atherosclerotic aneurysms (most common), foreign bodies, tumors, trauma, radiation therapy, arteritis, and infections.^[2] Secondary ArEF was first described by Brock in 1953 and occurs as a complication of aortic reconstructive surgery.^[1] Being far more common than primary ArEF, the secondary ArEF can be attributed to an increasing number of abdominal aortic repairs in the past decades.^[1]

The most common clinical symptoms of ArEF are abdominal pain and GI hemorrhage. GI hemorrhage may

start as self-limiting intermittent episodes often termed as herald bleeds, before massive bleeding with time intervals ranging from hours to months. This can be explained by local vasospasm and hypotension with the formation of a thrombus that may temporarily occlude the fistula. Later, when the effect of local vasospasm ceases and normotension is restored, the thrombus may dislodge and lead to massive bleeding. Other less common symptoms include pulsating abdominal mass, weight loss, fever, and sepsis.

IEF following radiotherapy appears to be an extremely rare entity with the first two cases reported by Kwon *et al.* in 1978^[3] (*radiation-induced AEF was reported before that date by Zarembok, 1972*). In a PubMed electronic literature search (conducted in September 2021) on “text word” synonyms for “arterioenteric fistula” and “iliac-enteric fistula” (limited to the English language), we have only found 12 case studies of radiation-induced IEF (excluded cases were describing IEF due to another etiology and AEF). These are summarized in [Table 1], including our present case.

The pathophysiology of IEF is not fully understood. It may develop as a result of chronic inflammation and accelerated atherosclerosis.^[4] It can also develop several years following radiotherapy.^[4] The majority of cases are primary IEFs with atherosclerotic aortoiliac aneurysms and prior non-vascular surgeries being the most common causes.^[4] The common iliac artery is the most frequently involved vessel.

CT angiogram is the most effective modality to detect the cause and location of GI tract bleeding. Besides active extravasation of contrast material into the bowel lumen, other typical imaging features include ectopic gas loculi adjacent to or within the involved vessel, obliteration of the fat plane between the affected vessel and bowel loop, disruption of vessel wall

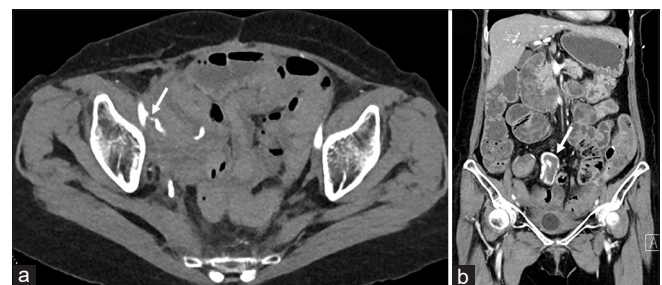


Figure 1: A 68-year-old woman with radiation-induced pelvic arterioenteric fistula presented with massive melena, hematochezia, and hematemesis. Contrast-enhanced CT (CECT) abdomen and pelvis, axial (a) image demonstrates a thin linear fistulous tract (arrow) between the medial wall of the right external iliac artery and the adjacent small bowel loop with contrast layering along the dependent wall of the bowel. Coronal (b) CECT image shows dilated small bowel loops with intraluminal hyperdense material and circumferential hyperdensity (arrow) in keeping with acute contrast extravasation outlining the wall of the small bowel.

Table 1: Summary of reported cases of radiation-induced iliac-enteric fistula in the literature.

Author	Fistula site	Radiation site/ neoplasm	Intervention	Outcome
Kwon <i>et al.</i> , 1978 ^[3] Reported two cases	Right IIA	Cervical cancer	Ligation	COB: Yes Survival: The patient died due to respiratory failure on POD 7
	Left CIA to ileum	Cervical cancer	Ligation	COB: Yes Survival: Yes
Vetto <i>et al.</i> , 1987 ^[4] Reported three cases	Left IIA to ileum	Rectal adenocarcinoma	Initial angioembolization	COB: Yes
	Left IIA to ileal conduit and jejunum	Rectal carcinoma	Rebleeding on the 3 rd day → Ligation Embolized with stainless steel coils Left axillofemoral bypass to restore leg perfusion	Survival: Yes COB: Yes Survival: Yes
	Left IIA to the sigmoid colon	Carcinoma of the bladder	Ligation.	COB: Yes Survival: Post-operative course was complicated by infections, abdominal abscesses, and respiratory failure. The patient died on the POD 58
Lukens <i>et al.</i> , 1995 ^[5]	Left IIA to the sigmoid colon	Cervical cancer	Not accessible	
Cohen <i>et al.</i> , 2017 ^[6]	Right internal iliac arteriovenous-enteric fistula	Rectal cancer	Embolization	COB: Yes Survival: Yes
Mikami <i>et al.</i> , 2017 ^[7] Reported two cases	Right EIA to the small intestine	Cervical cancer	Stent	COB: Yes, rebleeding after 3 weeks → coil embolization followed by f-f bypass Survival: Yes
	Right EIA to small bowel	Cervical cancer.	Embolization of the right iliac arteries and f-f bypass Aortic left CIA stent was placed to block blood flow into the right CIA Ligation of the distal right EIA to prevent backflow of blood through the f-f bypass	COB: Yes Survival: Yes
Mohammad, 2018 ^[8]	Right EIA to the small bowel	Urinary bladder carcinoma	Endovascular covered stent	COB: Yes Survival: Unknown
Dimech <i>et al.</i> , 2018 ^[2]	Right ileoduodenal fistula between the right CIA and duodenum	Ascending colon carcinoma	Endovascular covered stent	COB: Yes, massive rebleeding after 3 months Survival: The patient died shortly after rebleeding.
Kassamali <i>et al.</i> , 2019 ^[9] Indeterminate if secondary to tumor invasion or radiation-induced. Present case	Left CIA to adjacent small bowel	Colon and endometrial cancer	Stent	COB: Yes Survival: Yes
	Right EIA to adjacent distal ileal loops	Cervical cancer	Endovascular covered stent	COB: Yes Survival: Yes. Referred to vascular surgery for further management of fistula

CIA: Common iliac artery, COB: Cessation of bleeding, EIA: External iliac artery, f-f: Femorofemoral, IIA: Internal iliac artery, POD: Post-operative day

integrity, and focal bowel wall thickening.^[1] In the majority of IEF cases, there is often evidence of contrast extravasation into the bowel lumen. In contrast for cases of AEFs, contrast

extravasation into the bowel lumen is not a common feature on CT angiogram. Conventional catheter angiography is often required when the source of the hemorrhage is unclear.

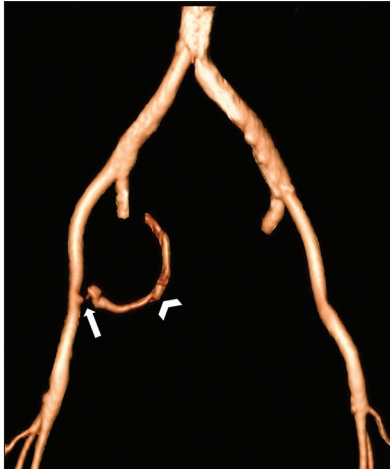


Figure 2: A 68-year-old woman with radiation-induced pelvic arterioenteric fistula presented with massive melena, hematochezia, and hematemesis. Three-dimensional volume rendering techniques of the contrast-enhanced CT angiogram with bone removal demonstrate active contrast extravasation from the medial wall of the right external iliac artery (arrow) with curvilinear streak (arrowhead) corresponding to the contrast outlining the wall of an adjacent loop of the small bowel.

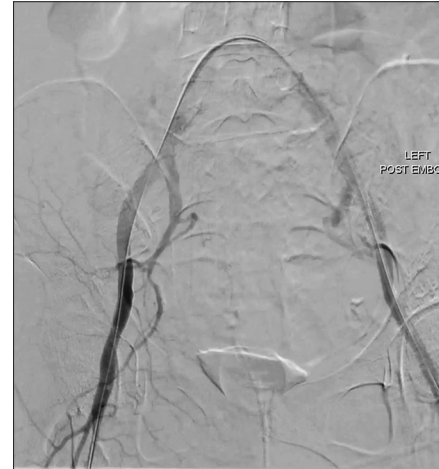


Figure 4: A 68-year-old woman with radiation-induced pelvic arterioenteric fistula presented with massive melena, hematochezia, and hematemesis. Digital subtraction angiography post-stenting angiogram cine images demonstrate cessation of contrast extravasation from the right external iliac artery.



Figure 3: A 68-year-old woman with radiation-induced pelvic arterioenteric fistula presented with massive melena, hematochezia, and hematemesis. Digital subtraction angiography initial angiogram cine images confirm active contrast extravasation from a thin linear fistula at the medial aspect of the right external iliac artery (arrow).

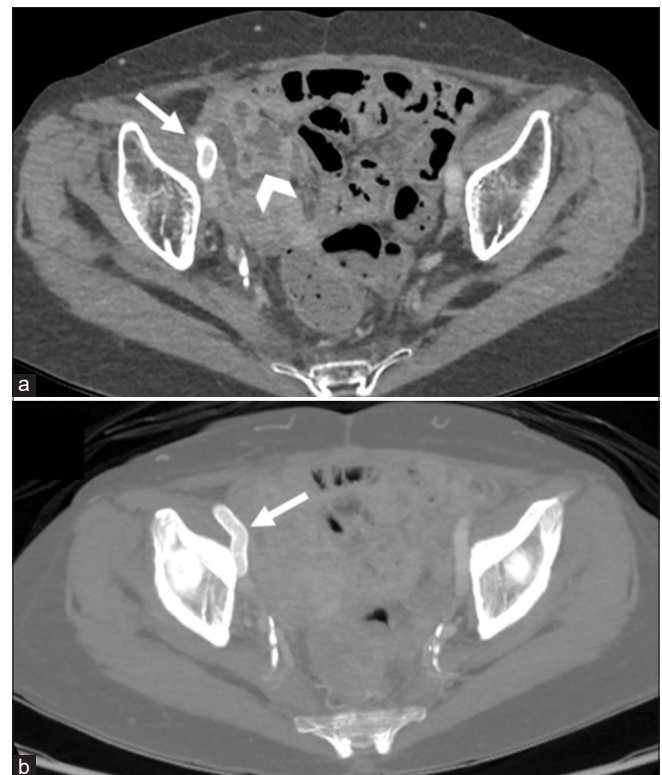


Figure 5: A 68-year-old woman with radiation-induced pelvic arterioenteric fistula was treated by the endovascular stent. Follow-up contrast-enhanced CT (CECT) abdomen and pelvis at 6 weeks after stenting. Axial (a) and maximum intensity projection (b) CECT images demonstrate the right EIA stent in situ with an adjacent loop of the small bowel (arrows). There is a mild mural thickening of the adjacent small bowel loop (arrowhead), which may be related to the prior radiotherapy. No evidence of active contrast extravasation or fistulation.



Video 1: A 68-year-old woman with radiation-induced pelvic arterioenteric fistula presented with massive melena, hematochezia, and hematemesis. Digital subtraction angiography initial angiogram cine images confirm active contrast extravasation from a thin linear fistula at the medial aspect of the right external iliac artery (arrow). Video is accessible from the portal.



Video 2: A 68-year-old woman with radiation-induced pelvic arterioenteric fistula presented with massive melena, hematochezia, and hematemesis. Digital subtraction angiography post-stenting angiogram cine images demonstrate cessation of contrast extravasation from the right external iliac artery. Video is accessible from the portal.

Due to its rarity, there is no consensus on the most appropriate management strategy for ArEF and IEF. The goals of treatment are to control hemorrhage, prevent a recurrence, and maintain adequate distal perfusion. The management strategy has evolved over the years. Conventionally, surgical repair was considered the only treatment. However, this approach was associated with significant mortality, particularly in hemodynamically unstable patients. In recent years, endovascular repair has proved to be a safe and effective technique for rapid control of bleeding from an ArEF.^[10] It

can be used as a temporizing treatment that provides time for hemodynamic stabilization, control of sepsis, and acts as a bridge to definitive surgical repair.^[10] Furthermore, it can be used as a long-term palliative treatment in patients who are at high risk for major surgery. Our case highlights the crucial role of endovascular repair in hemodynamically unstable patients presenting with massive hemorrhages due to an IEF. It also illustrates that endovascular repair can be used as a long-term palliative procedure.

CONCLUSION

Radiation-induced arterioenteric or IEF is a rare cause of severe lower GI bleeding which can potentially cause life-threatening complications. This condition should be suspected in patients with a prior history of pelvic radiotherapy. Prompt diagnosis facilitates early and appropriate treatment, especially when time is of the essence in an emergency setting. CT angiogram is the most effective imaging modality to diagnose GI hemorrhage and endovascular stent-graft placement is a safe and effective treatment.

Declaration of patient consent

Patient consent is not required as patients' identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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