

Case Report

DOI: <https://dx.doi.org/10.18203/2349-2902.ijssurgery20222243>

Primary aorto enteric fistula as a cause of massive upper gastro-intestinal bleed-a very rare case presentation

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Received: 03 August 2022

Accepted: 18 August 2022

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ABSTRACT

Primary aortoenteric fistula (PAEF) is spontaneous development of communication between the native aorta and anywhere within the gastrointestinal tract. It is extremely rare and fatal condition which usually presents as a painless upper gastrointestinal bleed. This condition is often overlooked because of its rarity and low index of suspicion by physicians despite the availability of a wide range of diagnostic tools. Computed tomography angiography (CTA) is the most common investigation done to diagnosis PAEF. This paper reports a case of 49 years old female with massive upper gastrointestinal (GI) bleeding. A PAEF was diagnosed by CTA which called for an emergency laparotomy with surgical repair of the fistula with a synthetic vascular bypass graft. The patient recovered well.

Keywords: PAEF, CTA, GI

INTRODUCTION

The overall incidence of aorto-enteric fistula (AEF) is 0.4-1.67%, of which primary AEF is found in 0.04-0.07% of all the cases in upper GI bleed.¹ PAEF is a spontaneous connection or fistulization between aorta and the alimentary tract. It occurs more commonly due to long standing extrinsic compression of an underlying aneurysm of the aorta that is unrecognized previously. It is more common in men than women with a ratio of 3:1.² The diagnosis is frequently delayed due to the rarity of the disease and low index of suspicion by physicians. Owing to the nonspecific and subtle clinical presentations, the diagnosis of AEF is difficult. The mortality rate associated with PAEF is very high almost 100% if treatment is delayed.³ A range of invasive and non-invasive diagnostic tools are available, but helical computer tomography (CT) remains the mainstay.

An aorto-duodenal fistula often presents itself with upper gastrointestinal bleeding and circulatory shock, which are symptoms identical to those of the much more common bleeding duodenal or gastric ulcers. Oesophago-gastro-duodenoscopy (OGD) is recommended as the first diagnostic choice in these cases and those having signs of upper gastrointestinal haemorrhage.⁴ Open surgery or endovascular aortic repair (EVAR) for AEF with bleeding has shown to improve the survival rate.

CASE REPORT

A 49-year-old female who is a known case of rheumatoid arthritis diagnosed 10 years ago and no previous history of any complaints presented with sudden onset of multiple episodes of hematemesis and melena for 3 days. There were no complaints of pain abdomen, vomiting or jaundice. On examination she was pale and tachycardic.

At the time of admission, the patient had blood pressure of 98/60 mm hg and pulse rate of 112/Mt and was afebrile. On abdominal examination she had mild tenderness in left lumbar region and per rectal examination showed tarry black stools. Blood investigations showed haemoglobin of 7 gm/dl with PCV-22 at the time of admission. Other blood investigations like liver function, kidney functions tests, platelet count and coagulation profile were within normal limits except for mild leucocytosis which showed 15200 /mm³. Upper GI endoscopy showed multiple blood clots in the stomach, duodenum and upper jejunum and any source of bleeding was not identified. CT angiogram showed 4.8×7.5×7 cm saccular abdominal aortic aneurysm (AAA) with possible communication at the level of D4 with eccentric thrombus and the left renal artery arising from the aneurysmal sac. She was planned for emergency laparotomy. Midline laparotomy was done and proceeded with limited Kocherisation. Intraoperatively pulsatile mass was palpable at the DJ flexure. Transverse colon attachments from spleen separated by dissecting the spleenocolic ligament. DJ flexure was found to be adherent to an aneurysmal sac of size 8×6×5 cm arising from just below the origin of superior mesenteric artery with extension towards left side. Left renal artery was arising from aorta with in the aneurysmal sac.

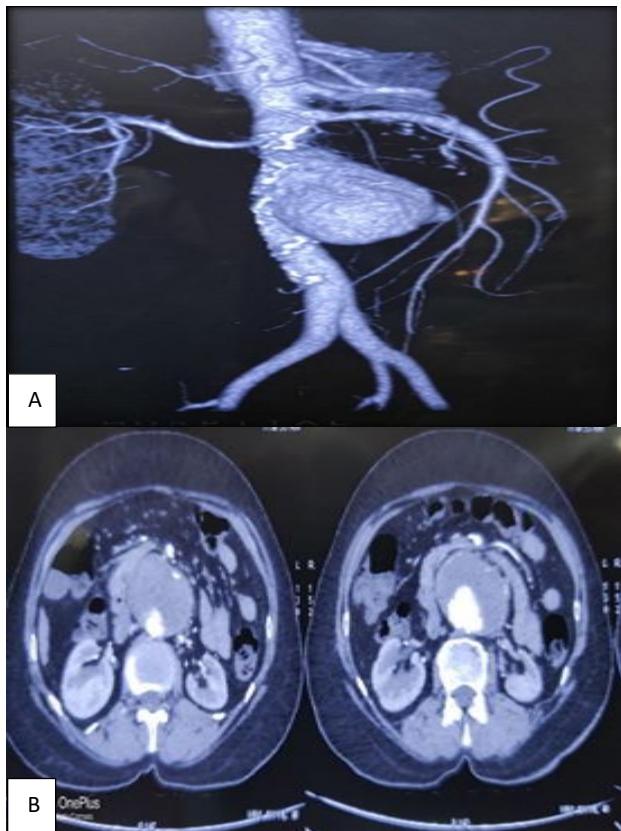


Figure 1 (A and B): CT abdominal angiogram. Large abdominal aorta saccular aneurysm 48×74×70 mm with eccentric thrombus with possible communication at D4 of duodenum.



Figure 2: CT abdominal angiogram. SMA originates from above the dilated segment. Left renal artery arising from within the thrombosed segment.



Figure 3: Intraoperative image showing aortic aneurysmal fistulazation with communication at D-J flexure.



Figure 4: After aneurysmorrhaphy and aortic and left renal bypass graft.

DJ flexure couldn't be separated from the aneurysmal sac, duodenum and proximal jejunum was transected with 80mm stapler. Left renal vein is retro aortic in position. Left renal vein and artery, right renal artery and vein controls were taken. Supraceliac and infra renal aortic controls were taken. Systemic heparinisation done and sac was opened after applying the clamp, and aortic thrombus was removed. Aortic tube graft with reconstruction of renal arteries bilaterally with same graft material of 6 mm size was done as bench preparation before getting in to surgical field, in view of reconstruction that may be need to aorta as well as renal arteries.

20 mm tube graft was anastomosed to aorta after SMA origin. Anterior to graft left renal artery bypass done with 6 mm Dacron graft. Right renal artery noted to be arising from the aorta just distal to the aneurysmal sac without communication. Distal anastomosis of the graft done to the infrarenal aorta and abdominal aortic aneuryssmorrhaphy, with aorto left renal bypass was done. Right renal artery reconstruction was not done. Gastrointestinal continuity maintained by anastomosing the previously transected proximal jejunum with duodenum at D2-D3 junction, in single layer with interrupted 3-0 vicryl sutures.

Post operatively, the patient had high Ryle's tube output from POD-2 until POD12. Aspirate was bilious in nature and amounted about 1.2 litres, 1.2 litre, 750 ml, 680 ml, 450 ml, 200 ml on POD 2, 4, 6, 8, 10 and 12 respectively which resolved with conservative management. This high output necessitated the need for total parenteral nutrition, orals clear liquids were started on POD-10 after intermittent blockage of Ryles tube and full oral diet was allowed from POD 13. Serial doppler ultrasound of abdomen was done in post operative period which

showed normal flow across the graft and normal flow along renal arteries with no e/o thrombosis and no intraabdominal collections. Renal function tests, Blood urea and S. creatinine in postoperative period showed increased parameters on day 1 and day 2-50/1.4 and 48/1.2 mg/dl and was in normal range since post operative day 3. She developed multiple parietal wall collections requiring ultrasound-guided aspiration-Clavien-Dindo grade-3a. Post operatively injection heparin 5000 IU IV was given every 6th hourly till post operative day 12 and dual antiplatelets were started after resuming oral diet. The aneurysmal sac culture was positive to MRSA (methicillin resistant *Staphylococcus aureus*) sensitive to linezolid and received it from POD 4 till pod 10.

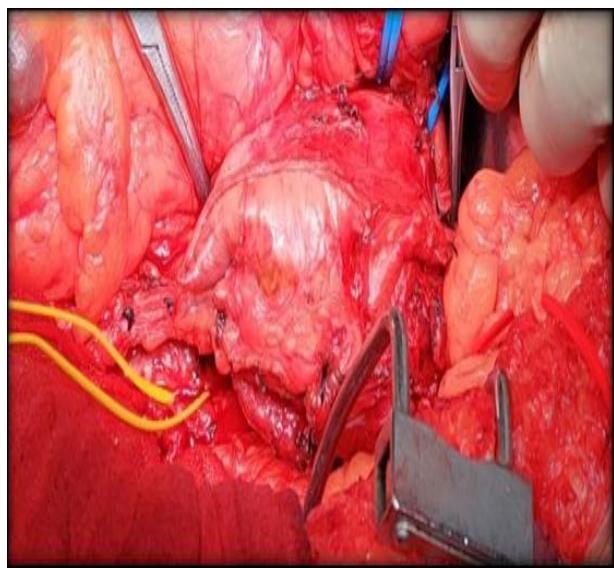


Figure 5: Aneurysmal sac with transected D-J flexure.

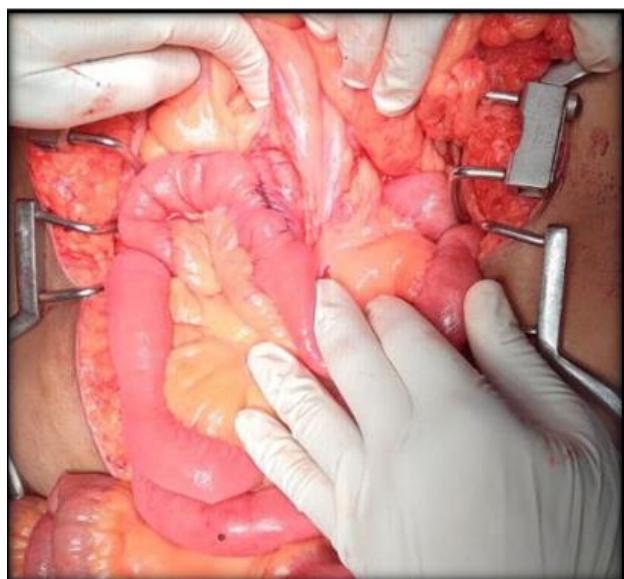


Figure 6: Gastro intestinal continuity by side-to-side proximal jejunal and duodenal (D2-D3 junction) anastomosis.

DISCUSSION

AEF is a rare condition of gastrointestinal bleeding with high mortality and is basically of two types PAEF and SAEF. Among which PAEF is rarest of the two and index of suspicion is very low compared to SAEF where there would already be a history of surgery or intervention to aorta. Suspicion of PAEF as a cause of upper GI bleed is very difficult especially with no previous history of any similar complainants in the past. A detailed history may sometimes reveal one or multiple episodes of sentinel bleed, which would have been spontaneously resolved without seeking any medical advise.

The incidence of secondary aortoenteric fistula has been on the rise since last two decades due to increased endovascular interventions and surgeries over aorta because of advancements in surgical and interventional methods. PAEF usually occur when there was a long standing unrepaired or unrecognized aortic aneurysmal sac that is free to erode into any adjacent gastrointestinal tract. PAEF may develop secondary to other autoimmune disorders or intrinsic wall defects of the vascular system. The fourth part of duodenum and duodenal jejunal flexure are in close contact with aorta at this location. The erosion of aneurysmal sac in to these parts of gastrointestinal tract is easy because of its close contact, due to relative fixity of duodenum and D-J flexure to the retroperitoneum, restricted mobility compared to other parts of gastrointestinal tract. Atherosclerosis is the most common cause which constitutes to 70-80%, others are infectious in 15% and rarely because of malignancy. Rarely, a PAEF is a result of cervical cancer, colon cancer, radiation therapy, diverticulitis, appendicitis, duodenal ulcers, cholecystitis, or foreign body.⁴ Pain, can occur in this condition due to increased size of the sac, active bleeding in to the lumen and sometimes an underlying infection with localized air pockets at the site of fistulisation. Pain is mostly localised to upper abdomen, back, or to the suprapubic region and depends on the location and extent of the aortoenteric fistula and should not be missed during history.

It was first described by Ashley Cooper in 1829 and the authorship of the term is attributed to him. The first case report was published by Salmon in 1843 and, in spite of the passage of one hundred and seventy years, there are to date only about 400 similar cases in the literature.⁵ AEFs occur in a male to female ratio of 3 to 1 and 8 to 1 in primary and secondary aortoenteric fistulas respectively.⁶ In a retrospective study conducted in 3,000 cases of bleeding above the ligament of trietz which was massive studied between January 1st 2007 to March 31st 2020 by Luo et al nine cases were only found to have AEF. Among which four had secondary aortoenteric fistula (SAEF) and five has PAEF. More than 50% of PAEF are situated in the duodenum near D-J flexure because of its close anatomical relation to the aorta. AEF develop most commonly in the lower third of the duodenum, due to its retroperitoneal location and

relatively fixated nature to the other retroperitoneal structures, including the aorta. Formation of AEF in other areas of the GIT is rare due to the mobility of the small and large intestine and the need for a fixated structure for the development of the fistula. Other locations include the oesophagus, stomach, ileum, jejunum, and colon have been reported but relatively rare.⁷ Cases of AEF in other locations have been also described, but are mostly secondary AEF and in the presence of previous graft and multiple adhesions. The classical symptoms of GI bleed, pulsating mass, pain was present in only 13.5% of cases as proposed by Scheltinga et al in a retrospective study.⁷ In PAEF aneurysm causes thinning of the aortic wall which rubs over the serosa of the small bowel with each pulsation of heart.⁸ This contact is more near the D-J flexure as it is the constant and fixed portion of small bowel allowing a more contact time with aorta as compared to rest of the bowel which undergo continuous peristalsis. A PAEF occurs in the setting of an unrepaired aortic aneurysm sac that erodes into the gastrointestinal lumen in the presence of predisposing factors.

Bleed and sepsis are devastating signs in AEF. Bleeding ranges from a small haemorrhage to overt lethal massive haemorrhage.⁹ CT with an oral contrast demonstrates an active extravasation of contrast in to aneurysmal sac or the vice versa. They showed that CT scans have an overall specificity of 100%, but the sensitivity was only 50%. Presence of air extraluminal air close to bowel wall or near-by an adjacent graft that was placed on a CT scan is almost pathognomonic of AEF. Because ulcers are common, OGD is the basic and first investigation for patients with upper gastrointestinal bleeding. An OGD identified the AEF or showed other abnormalities in only 25%-50% of confirmed AEF cases. Because typical OGD often omits the examination of distal duodenum and upper jejunum. Aortography does not recognize the fistula site exactly but if done may help in preoperative planning before and gives a road map before exploring. A retrospective study by Thompson that CT angiogram and endoscopy can identify a fistula in 35% and 25% of the cases respectively.¹⁰

Sepsis remains most dreaded complication in managing AEF if failed to manage mortality may reach more than 60%. As the management is often highly morbid due to surgery and high chance of sepsis, administration of I.V antibiotics should ideally cover both gram-negative, gram positive, anaerobic bacteria.¹¹ The goal of AEF repair are rapid and effective control of haemorrhage, preservation of perfusion to the legs and treatment of containment infection. Prompt treatment is essential. The mortality rate from surgery is 30-40% and surgical approach must consider the amount of retroperitoneal contamination, which was related to the bacterial flora and depending on the involved GI segment. There is no consensus on the choice of the surgical approach, because of lack of large case volumes or long-term studies. Extra-anatomical bypass and repair of the enteric tract is the treatment of choice in cases of gross contamination; however, this

technique is associated with survival rates of 40-60%, which was done in the present case study. Primary aortoenteric fistula with aneurysmal extension into left renal artery, requiring repair for the renal artery has not been reported in literature till date. This presentation is unique and was managed successfully even though there was increased postop morbidity she was discharged uneventfully on POD 14 and was on follow up since 10 months from date of surgery and was doing well.

CONCLUSION

Clinical presentation of the patients with AEF can take up to several hours; however, in fulminant presentation, death may occur within several minutes. So, only by placing PAEF top on the list of possible diagnosis, the condition could be recognized in due time. Early diagnosis by CT and timely surgical intervention are critical, though perioperative and late mortality are high especially in patients with positive bacterial cultures. Various minimally invasive interventional procedures have been proposed with success rates in literature, but till now surgery remains the single and best treatment modality for this fatal condition.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: Mudadla R, Kamal H, Anbalagan A, Purushothaman G, Satyanesan J. Primary aortoenteric fistula as a cause of massive upper gastro-intestinal bleed-a very rare case presentation. Int Surg J 2022;9:1635-9.