

Tuberculous aorto-duodenal fistula: a rare cause of upper gastrointestinal bleeding

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ABSTRACT

Aorto-enteric fistulas are rare and are associated with significant mortality. Infective causes usually occur within the setting of post-graft repair. Aorto-enteric fistula secondary to tuberculosis is extremely rare despite the high prevalence of this infection. Unfortunately, the diagnosis is often not suspected until surgery or at post-mortem. We report a case of an elderly Malay man presenting with massive gastrointestinal bleeding secondary to a tuberculous aorto-duodenal fistula in association with a saccular abdominal aortic aneurysm. This was successfully managed with an aortobifemoral graft repair and standard anti-tuberculous treatment for six months. A literature review of this rare condition is presented.

Keywords: aneurysm, fistula, gastrointestinal bleeding, tuberculosis

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INTRODUCTION

Aorto-enteric fistulas (AEFs) are rare and associated with high mortality.⁽¹⁾ Despite treatment, the mortality rate remains high. AEFs commonly occur within the setting of post aorto-iliac surgeries for aneurysms.⁽²⁾ Less common causes of AEFs include peptic ulcer diseases, malignancies, aortitis, previous radiation therapy and foreign bodies.⁽³⁾ The infective causes of AEFs are extremely rare; AEFs usually occur due to infection of the aortic grafts. Tuberculous causes of AEF are also extremely rare. Intermittent gastrointestinal bleeding (GIB) is a common presentation that can be obscure but is eventually massive.^(4,5) We report a case of a tuberculous AEF in association with a saccular abdominal aortic aneurysm presenting with massive GIB.

CASE REPORT

A 68-year-old Malay man presented with a three day history of central abdominal pain, melaena and a one day history of haematemesis. This was associated with dizzy

spells and syncope. The patient's past medical history included diabetes mellitus and pulmonary tuberculosis (TB) diagnosed three months earlier. He was still on standard anti-TB treatment and had been compliant with the treatment. Two weeks before the current presentation, he was referred for the evaluation of asymptomatic microcytic anaemia and positive faecal occult blood. The initial suspicion included the involvement of gastrointestinal TB. Upper gastrointestinal endoscopy (also known as oesophagogastroduodenoscopy [OGD]) revealed atrophic gastritis, and a colonoscopy revealed Grade II haemorrhoids. Ileoscopy did not reveal any TB involvement of the terminal ileum. Ultrasonography of the abdomen was normal.

On examination, the patient was pale but comfortable. He was hypotensive (87/41 mmHg) with a heart rate of 78/min. He had supraclavicular and cervical lymphadenopathy which was consistent with TB infection. Chest examination was unremarkable. The patient's abdomen was soft but tender on deep palpation in the epigastric and umbilical regions. There was also a pulsatile and expansile mass, measuring approximately 3.5 cm in diameter, which was consistent with an aneurysm. Digital rectal examination showed melaenic stools. The peripheral arterial pulses in the lower extremities were palpable but diminished considerably on the left side with an absent left dorsalis pedis pulse, but Doppler signals were present.

Laboratory investigations showed microcytic anaemia (haemoglobin 5.6, normal range [NR] 13.5–18.0 gm/dL; mean corpuscular volume 71.3, NR 80–99), slightly elevated urea (7.6, NR 2.5–6.4 mmol/L), hyperglycaemia (11.4, NR 3.3–6.1 mmol/L), marked hypoalbuminaemia (23, NR 35–48 gm/L) and an elevated erythrocyte sedimentation rate (108, NR < 10 mm/hr). The results of the rest of the investigations were within normal limits.

The patient was transfused with six units of packed cell blood and two units of fresh frozen plasma. An urgent OGD up to the level of the third part of the duodenum revealed blood clots in the stomach. Careful evaluation of the duodenum and stomach did not reveal any obvious

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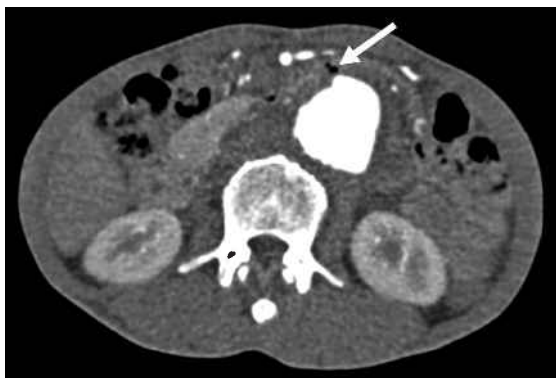


Fig. 1 Non-gastrointestinal contrast study shows ectopic gas collection (arrow) located in the aneurysm wall with an area of pointing within the aneurysm lumen, indicating the site of the fistula.

source of bleeding. A colonoscopy with ileoscopy showed melaenic stools. In view of the possibility of AEF, a referral was made to the vascular surgeon, and the intravenous administration of cefuroxime (1.5 g, three times a day) was started.

Computed tomography (CT) showed an infrarenal saccular aneurysm (3.5 cm) bulging towards the left. An intramural air pocket was seen in the aneurysm wall, which is suggestive of an AEF (Fig. 1). An urgent laparotomy was carried out, which confirmed an AEF between the fourth part of the duodenum and the saccular abdominal aortic aneurysm. There were also several enlarged (1 cm) para-aortic lymph nodes at the neck of the aneurysm, which was firmly adherent to the peritoneum. Upon opening the aneurysm sac after cross-clamping and evacuation of the mural clot, two aorto-duodenal fistulae (1.5 cm and < 1 cm) were seen. The aneurysm was repaired using an 18 Fr bifurcation Gortex graft, with the distal limbs anastomosed to the common iliac arteries. A segment of the duodenum containing the two fistulae was resected. Histology was positive for granulomatous changes and acid fast bacilli (Fig. 2).

Postoperatively, the patient's recovery was delayed due to small bowel obstruction secondary to ileus, which resolved with conservative treatment of intravenous fluid and resting of the gut. The patient was discharged on the 14th postoperative day. He was maintained on anti-TB treatment and had remained well on follow-up.

DISCUSSION

AEFs are connections between the aorta and bowel and are rare causes of GIB.⁽⁵⁾ AEFs have been categorised into two types. Secondary (Type II) AEFs are more common and are usually associated with previous aortic surgeries, especially abdominal aortic aneurysms graft repair.⁽²⁾ Primary (Type I) AEFs are usually associated

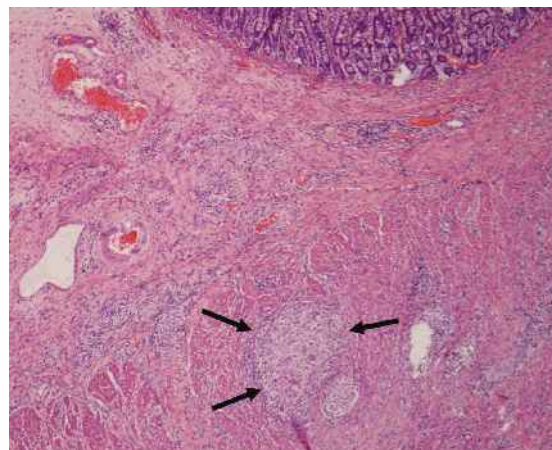


Fig. 2 Photomicrograph of a section of the affected duodenum shows granuloma (arrows) within the muscularis propria layer (Haematoxylin & eosin, $\times 4$).

with atherosclerotic aneurysms.⁽¹⁾

Infective causes are extremely rare and usually occur within the setting of an existing atherosclerotic aneurysm or post-graft repairs of the aneurysm. *Salmonella* is commonly implicated in infected native aneurysms, whereas *Staphylococcus epidermidis* and *Streptococcus faecalis* are commonly associated with post-repair graft infection.^(6,7) Despite a high prevalence of TB infections, tuberculous AEFs are extremely rare. There have been less than 50 cases of TB aneurysms reported to date.⁽⁸⁾ Both thoracic and abdominal aortas are equally affected, with most aneurysms being saccular (90%) and of the false type (88%).⁽⁸⁾ Disseminated TB was present in 46% of the cases.⁽⁸⁾ Only six cases of tuberculous aorto-duodenal fistula have been reported in the English literature (Table I).^(4,9-13) The first case was a secondary tuberculous aorto-duodenal fistula reported in 1968. The fistula developed following a surgical graft repair of a ruptured abdominal aortic aneurysm, resulting in a massive fatal GIB.⁽⁹⁾ The duodenum is the most commonly affected site; the relatively fixed position being the third part of the duodenum, as it crosses the aorta, which probably accounts for the predilection for this site.⁽¹⁴⁾

Four types of tuberculous arterial involvement have been described, including Type 1: miliary TB of the intima, Type 2: polypoidal or vegetations containing tuberculous tissue attached to the intima, Type 3: TB involvement of several layers of the vessel walls, and Type 4: tuberculous aneurysms which are typically saccular in nature.⁽⁸⁾ Our patient had Type 4 involvement, which is the most commonly reported manifestation.

The underlying pathogenesis of formation of tuberculous AEFs can occur via three pathways. The most common pathway reported in the literature is by

Table 1. Reported cases of aorto-duodenal fistulas secondary to tuberculosis infection.

Case	Age/ gender	Presentation (GIB)	History of TB	EGD (AEF)	Aneurysm	Site of AEF (Duodenum)	Diagnosis	Treatment	Outcome
Eadie et al ⁽⁹⁾	80/M	Recurrent	Previous PTB	Barium study	Infra-renal	3rd part	Surgery (10 mm)*	Resection / repair	Died
Goldbaum et al ⁽¹⁰⁾	75/M	Massive	Previous PTB	Not seen	No (Friable aorta)	3rd part	Surgery	Y-shaped Dacron graft	Survived
Allins et al ⁽¹¹⁾	77/M	Massive	None	Not done	Pseudo aneurysm	4th part	Surgery (15 mm)*	Repair/ axillio- femoral bypass	Died
de Kruijf et al ⁽¹²⁾	38/M	Recurrent	Current TB LN	Not seen	Saccular (30 mm)	3rd part	Surgery (1 mm)*	Aneurysmectomy	Survived
Tsai et al ⁽¹³⁾	80/M	Recurrent	Current PTB	Not seen	Saccular	3rd part	Surgery	Resection/ Dacron (18 mm) graft	Died
Tsai et al ⁽⁴⁾	69/M	Recurrent	Previous PTB	Seen (not suspected)	Pseudo aneurysm	3rd part	Surgery	Dacron graft	Survived
Present case	68/M	Recurrent / Massive	Current PTB/ TB LN	Not seen	Saccular (35 mm)	4th part	Surgery	Gortex graft	Survived

*size of fistula

AEF: aorto-enteric fistula; EGD: esophago-gastroduodenoscopy; GIB: gastrointestinal bleeding; TB: tuberculosis; PTB: pulmonary tuberculosis; LN: lymph adenopathy; M: male

direct extension from the adjacent infected organs such as the para-aortic lymph nodes, pericarditis, empyema, spondylitis or paravertebral abscess, compared to the other two pathways which are either by direct spread through a defective aortic intima or by haematogenous spread to the vasa vasorum.⁽⁸⁾ In one review, the presence of contagious organs could be detected in up to 75% of the cases.⁽⁸⁾ The presence of multiple para-aortic lymph nodes in our patient suggests that this may be the most likely pathogenesis.

The clinical manifestations of tuberculous AEFs are similar to those of AEFs of other aetiologies. Non-specific symptoms of TB infections may be present. Clinical presentations of any AEF include persistent abdominal pain, bleeding or an abdominal mass that is either palpable or radiologically detectable. This classical triad is seen in only 6%–27% of patients with non-tuberculous AEFs.⁽¹⁴⁾ GIB may be initially mild and obscure (herald bleed), and is reported to be the initial presentation in over 94% of AEFs, regardless of the aetiology.⁽¹⁵⁾ GIB is often eventually catastrophic. This interval has been reported to range from five hours to five months with a median of four days.⁽¹⁴⁾ TB involvement in other organs has been reported in up to 63% of cases of a TB-associated aneurysm, which provides important clues.⁽⁸⁾

AEFs are associated with very high mortality, approaching 100%, if untreated. Unfortunately, despite treatment, the reported mortality rate remains high (10%–70%). Therefore, management requires a high index of suspicion. In our case, the diagnosis was suspected early before surgery. However, in most reported cases, the diagnoses were only made during surgery or at post-mortem.

An endoscopy is important to exclude other causes of GIB. However, AEFs are seldom visualised or detected during endoscopy. If AEFs are suspected, endoscopic evaluation should be avoided due to the associated risk of uncontrollable bleeding. Other investigations such as CT imaging, angiography, and radionuclide scintigraphy often fail to show the fistula tract unless there is active bleeding.^(16,17) CT imaging is important, and findings, such as active contrast extravasations into the adjacent bowel, ectopic gas and bowel thickening, are suggestive of AEF. In our patient, the finding of the air pocket was diagnostic. However, it is important to consider AEF in any patient with an aneurysm presenting with GIB, regardless of the size of the aneurysm and even in the absence of any of the suggestive findings described above.

The management of tuberculous AEFs is similar to that of AEFs of other aetiologies, except that in cases of tuberculous AEFs, anti-TB therapy is mandatory. Surgery should be considered early in all suspected cases. Surgical graft repairs or endovascular stenting are available options.⁽⁵⁾ A review of tuberculous mycotic aneurysms of the aorta showed that treatments with surgical or medical therapies alone were associated with a high mortality rate, whereas all patients receiving combination therapies survived.⁽⁸⁾

In conclusion, the diagnosis of AEF should be considered in patients presenting with GIB associated with an aortic aneurysm. Failure or a delay in diagnosis is often associated with significant mortality. Early surgical repair and replacement of the aneurysm with bowel resection is the preferred treatment option, and in tuberculous AEFs, this should be followed by a complete course of anti-TB treatment.

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